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HYPOSPADIAS

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IN hypospadias one can find the abnormal opening of the urethra at any point in the midline of the ventral surface of the penis from the frenal position to the peno-scrotal junction, in the midline of the scrotum or in the perineum. Chordee in greater or less degree is an almost invariable accompaniment of all types of hypospadias. A third deformity is the characteristic loose hood-like prepuce.

Surgical cure of hypospadias is required only if, because of his disability, the patient—

(i) cannot pass urine standing up, without dribbling or soiling his clothes, or

(ii) will be unable to co-habit or procreate normally when he reaches man's estate, or

(iii) has a degree of deformity which, while not as gross as in (i) or (ii) is sufficiently abnormal to make him unduly self-conscious or to subject him to unpleasant attention or even ridicule, first, at school, and later, in adult life.

It is generally accepted that minor degrees of hypospadias, that is to say, cases with the urethral orifice in the region of the frenal position or even somewhat further back, require no major surgery, provided the penis is almost straight and has adequate ventral length; this holds good even if there is a degree of chordee, provided this element of chordee is strictly limited to the glans and does not involve the body of the penis at all.

Indeed, as Twistington Higgins, Williams and Nash (1951) very rightly point out, attempted urethral repair in such cases may interfere with local sensitivity to such an extent as to render the operation positively harmful. Once it has been decided that plastic operative repair of a frenal type hypospadias is unnecessary the excessive tissue in the hooded prepuce may be excised to improve the appearance of the penis; sometimes also in these cases a simple meatotomy to provide an adequate urethral orifice is found necessary. Any further interference in these cases can only be described as meddlesome surgery.

Mid-penile cases can be equivocal, in as much as the decision for or against surgery generally rests on the degree of chordee which is present; as a generalization, one can say that operation is, as a rule, indicated in these cases. With a urethral opening further back than this point, surgery is invariably needed.

Many cases of hypospadias with marked chordee have the urethral orifice apparently well forward on the under surface of the penis, even actually quite close to, or even at, the frenal position but the gross ventral bowing of the penis demands correction; in these cases, when the penis is properly straightened by surgery and given adequate ventral length, the urethral orifice is, at the same time, inevitably displaced back to the region of the peno-scrotal junction, and this necessitates subsequent reconstruction of the penile urethra.

When no testes are in the scrotum, the possibility of intersex, in what appears at

first sight to be a simple case of hypospadias, should always be remembered.

If doubt exists a careful clinical examination, including the use of an infants' cystoscope, estimation of 17-ketosteroids and a skin biopsy will help to decide the gonadal sex; in some circumstances a laparotomy and gonadal biopsy will be indicated.

In male intersex, when the victim is lucky enough to have a hypospadiac penis—instead of presenting with completely feminized external genitalia—repair of the hypospadias is usually possible though, as Russell (1954) remarks, phallic hypoplasia and chordee are often most marked in such cases.

On the other hand female intersex of the adrenogenital pattern, which, in my experience, is much commoner than male intersex, usually presents with a hypospadiac phallus much larger than occurs with male intersex; indeed, as Williams (1954) points out, it may be larger than the penis of some boys of the same age. In the rare case where the labio-scrotal folds have fused, the likeness to a straightforward male hypospadias is most confusing. However, it is very rarely indeed that, in such a case, circumstances warrant masculinization by repair of the hypospadiac phallus rather than feminization with the use of cortisone, aided by appropriate plastic surgery.

I think most surgeons of experience will agree that surgical repair of hypospadias presents some very special problems from the point of view of uninterrupted recovery, limitation of the hospitalization period, and satisfactory anatomical and functional end-results.

If any evidence is needed to fortify this opinion one has only to turn to the many and varied techniques in this particular branch of surgery which have been extolled from time to time in the past, only to be subsequently discarded. According to Campbell (1951) more than 150 operative techniques for the correction of hypospadias have been described.

I have often been struck by the exaggerated claims made for various methods of surgical attack; quite often however, the successful cases referred to are few in number, or else a larger series of cases is spoken of in general terms, but failures and delays and persisting post-operative fistulae are glossed over or are certainly not recorded in full detail.

Cecil (1932) wrote a comprehensive account of the many methods of hypospadias repair which had appeared in the literature up to that time. Since then considerable strides have been made and the methods practised in most large centres are now limited to three or four different techniques with, of course, the inevitable minor variations adopted by individual surgeons.

Over the last twenty-five years I have had many setbacks with cure of hypospadias but the wealth of material available in the last eleven years has enabled me to make a critical survey of the end-results of different methods.

Some seven years ago I reported five cases (Fraser, 1950) which I had recently operated on, employing suprapubic drainage and keeping the patient in hospital until a two-stage operation had been completed using a technique modelled on that used by Cabot (1936). The sound idea of burying the newly formed urethra deeply, by anchoring the penis in the clitoridis position, which is the basic principle in this operation, was introduced by Bucknall in 1907. Unfortunately, as he was the first to admit, his technique involved the use of scrotal hair-bearing skin in the formation of the new urethral tube, a fault which is eliminated in the Cabot operation. As late as 1936, Walters, reviewing the literature of hypospadias repair spoke favourably of the use of the Bucknall operation, while admitting the possibility of hair growth and calculus formation in the new urethra. Due credit must be paid to Bucknall for his originality and vision in introducing a surgical technique which, in general principles, has stood the test of time for fifty years and which is, with certain specific modifications, still as popular as any other method now in use.

On the whole the results of this small series in 1950 using the Cabot technique were very satisfactory but the stay in hospital was unduly prolonged and there was danger of sloughing of the flaps if these were not "delayed" before the final stage.

This led to a trial series using the method described by Denis Browne (1949), which is based on a principle used by Hamilton

Russell (1923) of Melbourne for cure of urethral stricture and first suggested indirectly by Duplay in 1880. In the Denis Browne operation the perineal urethrostomy used in place of the suprapubic cystostomy to by-pass the urine was found to be satisfactory and comfortable for the patient. The operative repair itself is simple, and the hospitalization time in a successful case is satisfactorily short. However, although the method has some enthusiastic supporters including Burns (1950), it has been appraised somewhat critically by Gross (1953), Cecil (1955), Crevy (1955) and McCollum *et alii* (1956).

It is an elementary principle of plastic surgery that scarring and cicatrization of a raw surface is minimized immediately it is covered with epithelium. In the Denis Browne operation however, some time is taken for epithelium to work its way round each side of the new urethra over a raw area of tissue; under these circumstances one would expect some scarring and subsequent cicatrization with narrowing of the lumen of the new urethral. The experimental work of Nesbit *et alii* (1950) however suggests that the subsequent fibrosis in this situation may not be as extreme as one might suppose, and Browne (1953) presents evidence to support his contention that serious stricture of the new urethra is not likely to occur.

In spite then, of the disregard of a generally accepted surgical principle, it is doubtful if any evidence in favour of serious stricture formation is yet proven and it is not on this premise that the method is really open to criticism.

A cosmetic weakness in the operation is that the scrotal skin which is dragged up on to the penis, tends with the passage of time to drag the penis down and does not allow of a definitive peno-scrotal angle. This however, does not result in any functional disability.

Whatever else may be said for or against the Denis Browne operation, it is incontrovertible that its crucial weakness lies in the liability to fistula formation. This tendency to fistula formation is understandable when one considers the small margin of safety which the operation provides. The epithelium which has to grow round to line the roof of the new urethra, is in juxta-position to the ventral skin surface of the penis, and any failure at all of perfect skin union on the ventral surface of the penis quickly results in the formation of a short epithelial-lined track from the new urethra to the surface, with the inevitability of a persisting fistula: for the same reason, a stitch hole may quickly become epithelized, again with the formation of a persisting fistula to one or other side of the midline of the ventral surface of the penis.

Certainly in my hands, the number of persisting post-operative fistulae following the Denis Browne operation was far too high to be pleasant and the method was discarded.

At this stage I decided to revert, in principle, to the Cabot operation with certain modifications calculated to make the operation safer and the hospitalization period shorter.

A perineal urethrostomy was found to have certain advantages over the suprapubic route to by-pass the urine and was persisted with; it is simple to perform and obviates the occasional leak of urine through the newly formed urethra which occurs with suprapubic drainage.

Instead of keeping the patient in hospital until the two stages of the operation were completed, necessitating long continued recumbency, the operation was done in two short stages, with an interval of three to four months between stages, during which time the patient was ambulatory and able to return happily to school. The technique employed followed in broad outline that advocated by Cecil in 1946 and 1955 and by Culp in 1951. The opportunity was also taken to introduce an innovation suggested by a useful step in the Denis Browne operation, in order to make more certain of perfectly sound healing of the wound with no possibility of breakdown after the first stage operation.

It is generally accepted that the only proper way to satisfactorily assess any surgical technique is to carry out that technique without deviation in a sufficiently large number of consecutive cases; only in this way can the advantages and disadvantages of any given method be presented in their proper perspective.

I therefore proceeded to use this method in a consecutive series of 9 cases which apart from the preliminary cures of chordee some years previously, were commenced and completed over the period 13th July, 1956, to the 12th September, 1957. The case histories were carefully tabulated and the course of events in each case meticulously recorded, with particular reference to any delays or unfavourable post-operative features, so that there would be definitive statistical data which would give a reliable guide to the merits or demerits of the method under review.

Over the period under review a number of cases already started with different techniques were completed and a number of others had the preliminary operation for cure of chordee; all of these cases naturally enough, have also been excluded from the series.

The method to be described is applicable to all cases in which, after the penis has been straightened, the urethral orifice is at or near the perineal urethrostomy wound from which the catheter had been removed.

TABLE 1

DETAILS OF THE CURE OF CHORDEE AND OF THE SUBSEQUENT ADJUSTMENTS FOUND NECESSARY PRIOR TO THE DEFINITIVE HYPOSPADIAS REPAIR

Patient	*Age at Operation for Cure of Chordee*	*Type of Operation for Cure of Chordee*	*Subsequent Adjustments Prior to Definitive Hypospadias Repair*
I.T.	6½ years	Preputial strap flap	Meatotomy and excision of excess ventral skin.
K.F.	5 years	Preputial strap flap	——
R.W.	4 years	Preputial strap flap	Meatotomy and excision of excess ventral skin.
K.C.	3½ years	Preputial strap flap	Meatotomy and secondary chordee repair (Byars operation). Secondary meatotomy.
R.H.	3½ years	Preputial strap flap	——
S.McL.	3 years	Preputial strap flap	Meatotomy.
K.R.	4½ years	Byars Operation	——
N.W.	2½ years	Preputial strap flap	Meatotomy.
A.S.	5½ years	Preputial strap flap	——

The only hypospadiac repair commenced and completed over this period which was not included in the series, was one in which, early in the period under consideration, trouble was experienced with the Malecot catheter in the perineal urethrostomy wound. This catheter obstructed in the first twenty-four hours after operation; a suprapubic cystostomy was then done and the case was successfully completed by the same technique but with one long stay in hospital rather than with two short stays. Later experience has shown that the offending Malecot catheter could have been removed and the case handled like all the others, with satisfactory temporary by-passing of the urine through the peno-scrotal junction or even some distance back on the scrotum. The method can also be used for perineal hypospadias but this requires an additional operation to bring the urethral orifice up to the peno-scrotal junction. In my experience perineal hypospadias is very rare compared with the other types and when it does occur, successful closure up to the peno-scrotal junction offers little technical difficulty unless there is a complicating recto-urethral fistula.

It will be appreciated that prior to the actual urethral reconstruction in each of the 9 cases in the series, the penis had already been satisfactorily straightened by previous

surgery, and the urethral orifice was of adequate calibre and was situated at or near the peno-scrotal junction.

As a matter of interest all but one of the nine patients had the chordee cured by the simple strap flap operation (Table 1) and the technique employed in the preliminary cure of chordee in eight of the nine cases has been included for completeness.

CURE OF CHORDEE

In this operation the surgeon must set out to remove all the constricting fibrous remnants of the urethral cord and corpus spongiosum on the under surface of the penis, which cause the ventral bowing. This may be undertaken at any age from two to five years depending on the size of the penis. Sometimes of course, patients do not present until they are considerably older.

A transverse incision is made on the ventral surface of the penis at the point of greatest bowing; all the fibrous elements on the under surface of the corpora cavernosa are painstakingly removed; to do this adequately, particularly in those cases where there is extreme bowing and a urethral orifice apparently well forward, one has to undermine the urethra, which becomes displaced backwards towards the peno-scrotal junction as the penis straightens progressively with the removal of the constricting elements. This dissection is not complete until the penis can be hyper-extended easily over the left index finger. This leaves a raw diamond-shaped area on the under-surface of the penis. A transverse incision is then made on the dorsum of the penis along the prepuce 3 mm. behind its coronal attachment. The two layers of the prepuce are then dissected free from each other and a longitudinal incision through the reflected layer on each side allows one to unfold the prepuce as a rectangular flap over the top of the glans. This flap is thin but its viability is amazing.

After the narrow isthmus of skin on each side of the penis has been divided in order to connect the raw area on the dorsum with the raw area on the ventral surface, the head of the penis is passed through a transverse slit made in the preputial flap. The segment of the flap distal to the slit is then sutured snugly into position over the raw surface on the ventral aspect of the penis, discarding any preputial tissue in excess of that required to cover the raw area smoothly and without tension. The proximal edge of the slit is then sutured to the fringe of prepuce remaining on the dorsum. An in-dwelling catheter is sutured in position and an occlusive dressing of tulle gras, gauze and adhesive strapping is anchored around the penis to minimize post-operative oedema.

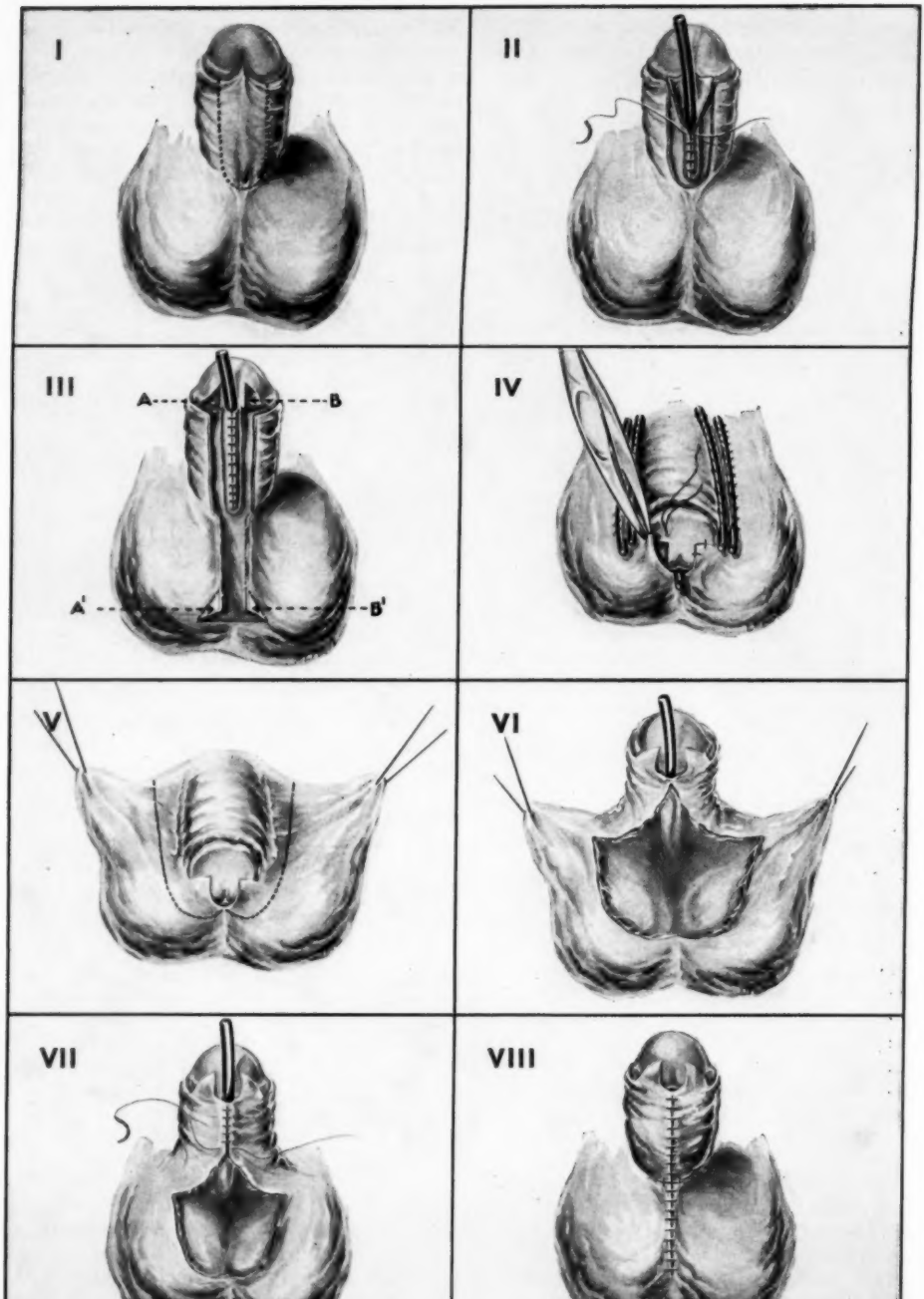
Alternatively Edmunds' (1926) method of using preputial skin in the correction of chordee still has many adherents and Byar's (1955) technique provides yet another way in which the preputial skin can be used to cover the raw ventral surface of the penis during the cure of chordee.

Many other methods of straightening the penis may be employed, but the use of a preputial flap provides adequate elastic non-hair-bearing skin on the ventral aspect of the penis, which will be valuable, subsequently, to form the new urethra. Matthews (1952) makes the point that the skin of the prepuce is histologically very like the normal urethral lining and this makes it particularly suitable for fashioning the urethra.

The criticism that the strap-flap method of straightening tends to leave the patient with redundant loose skin on the under-surface of the penis is of no real import provided care is taken to ensure that the preputial flap used is smooth and is trimmed down to the correct size and shape. Mays (1951) has pointed out that the elastic skin of a ventral preputial flap settles well into position and develops a very ample blood supply.

In preparing the patient for the hypospadias repair proper it is not only necessary to have a straight penis with smooth non-hair-bearing ventral skin, but also to ensure that the existing urethral orifice is of adequate calibre: to this end a simple meatotomy is sometimes necessary (Table 1).

Any method of chordee repair which utilizes skin from the sides or under-surface of the penis prohibits the use of this skin for the subsequent formation of the urethra, the reason being that careful examination of the average adult penis will show that this skin always grows hair in greater or less degree; if this skin is used to fashion the new urethra it will contain hair-bearing



Captions at foot of opposite page.

follicles which will inevitably become active and produce hairs in the urethral lumen with subsequent complications which are troublesome and often difficult to cope with.

For all these reasons the practice of making a transverse incision on the ventral aspect of the penis, excising the subjacent tissue and simply sewing the wound up longitudinally is an unsatisfactory method of chordee repair especially if this ventral penile skin is to be used for any part of the subsequent urethral reconstruction.

It is obvious that adequate straightening of the penis is a necessary prerequisite to urethral reconstruction. Occasionally bowing will not be overcome at the first attempt and a second operation becomes necessary; this occurred in one of the nine cases under review.

THE HYPOSPADIAS REPAIR

Photographs taken during the course of the operation in an effort to explain the technique were unsatisfactory: following this, close examination of a number of papers, showing photographs of other operations, convinced me that sketches made in the theatre and subsequently elaborated into coloured drawings, help to demonstrate the various stages of an operation far better than clinical photography in the theatre. Therefore in describing the technique employed, drawings are presented in preference to photographs; photography however, is obviously the only satisfactory way to provide factual evidence of the end-results in individual cases.

As soon as the penis and scrotum are of adequate size one can proceed with the repair proper. The age will vary in individuals but in the average case this operation can be undertaken at five or six years old.

First stage

A Malecot catheter is passed into the bladder on a sound small enough to allow it to slide easily inside the catheter. The sound is withdrawn to make sure that the urine is flowing freely through the catheter. The sound is then re-introduced into the catheter and turned so that its point projects up in the midline of the perineum. With a diathermy knife a cut is made down to the catheter at the spot where the tip of the sound is projecting it towards the surface — care being taken to keep in the midline — until the rubber of the catheter is exposed; the catheter is then seized, the sound is withdrawn and the distal end of the catheter is pulled back through the urethra until it is brought out through the perineal wound. The catheter is then anchored by a rubber cuff to the perincal skin, first ensuring that the wings of the end of the Malecot catheter are not pressing on and irritating the internal urethral orifice. Effective and comfortable by-passing of the urine from the operation site is thus effected. Some surgeons prefer a Foley to a Malecot catheter but this seems to be a matter of individual choice. While a diathermy knife is useful for the cut down on to the catheter this manoeuvre can quite well be carried out by using an ordinary fine bladed scalpel.

The surgeon then proceeds with the urethral reconstruction.

A Duplay flap is mapped out on the ventral surface of the penis encircling the existing urethral orifice and passing forward on each side right up to the glans (Fig. I). Care is taken to see that the flap is made wide enough to provide an adequate urethral lumen. This flap is partially freed on each side and the edges inverted and united over a rubber

FIG. I. The Duplay flap outlined for the commencement of the first stage of the hypospadias repair.

FIG. II. The new urethra being established.

FIG. III. The new urethra completed and the scrotal incision made. The triangular areas on the glans, A and B, are made raw to receive the triangular scrotal skin flaps A^1 and B^1, when the penis is ventroflexed on to the scrotum.

FIG. IV. The penis sutured into position on the scrotum, with the attachment of the triangular scrotal flaps on to the glans in process of completion. Note the continuous line of coaptation of the opposed raw surfaces on each side, achieved by the use of rubber tubes held in position with a single layer of interrupted vertical mattress sutures.

FIG. V. The scrotal flaps outlined for the commencement of the second stage of the hypospadias repair.

FIG. VI. The penis, together with the new urethra and the borrowed scrotal skin, lifted from the scrotal bed.

FIG. VII. The ventral surface of the penis being closed over the preformed urethra.

FIG. VIII. Completion of closure of the ventral surface of the penis and the gap in the scrotum.

catheter to form the new urethra (Fig. II). The catheter varies in size with the age of the patient and the size of the penis; this catheter passes down only to just beyond the commencement of the new urethra. It acts purely as a scaffolding over which the urethra is constructed without any tension and is left *in situ* after the operation; no harm has accrued from this. Suturing is done with interrupted 0000 catgut sutures, using an eyeless needle. Care is taken to see that the corium only is picked up with the needle.

A longitudinal midline incision is then made in the scrotum slightly longer than the length of the penis. At the extremity of this incision a small transverse cut is made and the longitudinal incision is slightly undermined on each side to allow the edges to be everted (Fig. III).

The penis is then folded ventrally on to the scrotum and the raw edges of skin on the respective sides of the penis and scrotum are approximated with interrupted sutures, utilizing two fine rubber tubes to ensure that there is a continuous broad area of coaptation of the respective raw surfaces on each side.

In doing this care is taken to anchor a triangle of scrotal skin to a rawed area on each side of the glans especially prepared to receive it (Fig. IV). This is the important addition to the Cecil technique, mentioned previously, which effectively prevents the glans penis from lifting away from the scrotum during the healing stage even if there is troublesome oedema or haematoma formation following the operation. The edge of the transverse cut made at the extremity of the scrotal incision will then lie snugly against the distal extremity of the new urethra to which it is anchored with several interrupted sutures. This is a further safeguard against any breakdown at this vital point during the healing stage.

The penis is thus firmly anchored to the scrotum in the clitoridis position with a new urethra already formed and deeply buried in the scrotal tissues. This is the whole crux of the operation.

The Malecot catheter is closed with a small spigot and during the post-operative period it is drained into a kidney tray every $1\frac{1}{2}$ hours. This provides a simple, painless and effective by-passing of the urine during the healing stage.

The operation area is left exposed and in the post-operative period care is taken to prevent any exudation accumulating on the suture lines; the foot of the bed is elevated for four days.

The amount of post-operative oedema varies but in the majority of cases it is much less marked than one would expect.

All the sutures, the small rubber catheter used as a scaffolding on the new urethra, and the Malecot catheter in the perineal urethrostomy wound, are all removed in seven days. The perineal wound nearly always closes quickly and the patient is ready for discharge within a fortnight after admission, happily passing his urine in the standing position, through his new urethra. He goes home for three or four months by which time all oedema has subsided and very little evidence of the operation remains, apart from the anchored down appearance of the penis.

Second stage

An incision is carried down each side of the scrotum. These incisions curve medially to meet in the midline just below the opening of the new urethra near the tip of the penis (Fig. V). The scrotal flaps are dissected up towards the midline; a catheter in the new urethra allows the urethra to be lifted safely off the scrotum with the flaps (Fig. VI). The two small scrotal flaps are then united on the ventral surface of the penis over the already well established new urethra (Fig. VII).

The closure of the gap left on the scrotum is easily effected and results in the production of a well-defined angle at the peno-scrotal junction (Fig. VIII). Very little post-operative swelling occurs as there is no extensive undermining of flaps. The wound is again left exposed and kept free of exudation; it heals quickly. A catheter is left *in situ* and is removed on the fourth post-operative day.

Anaesthetic technique

Adequate pre-operative sedation is particularly important for children undergoing repair of hypospadias. They are nervous and easily upset, and become more difficult to manage with each anaesthetic they receive.

Pentobarbital is given to all cases on the night before operation. For premedication, children of seven years and under have pentobarbital and atropine; older children receive papaveretum and scopolamine with a small dose of pentobarbital.

Induction of anaesthesia, when possible, is by intravenous thiopentone. All cases have been intubated with the aid of a muscle relaxant, and anaesthesia is maintained with endotracheal nitrous oxide, oxygen and ether, using a T-piece assembly with minimal rebreathing. Ether is not introduced until the use of diathermy for the urethrostomy has been completed.

Induced hypotension, using trimetaphan has become part of the technique for Stage 1 of the hypospadias repair proper. Continuous oozing of blood had always been a hindrance in this stage until induced hypotension was introduced for this series of cases. Blood loss is now minimal, the operative field remains clear and there has been a marked reduction in post-operative oedema of the penis and scrotum.

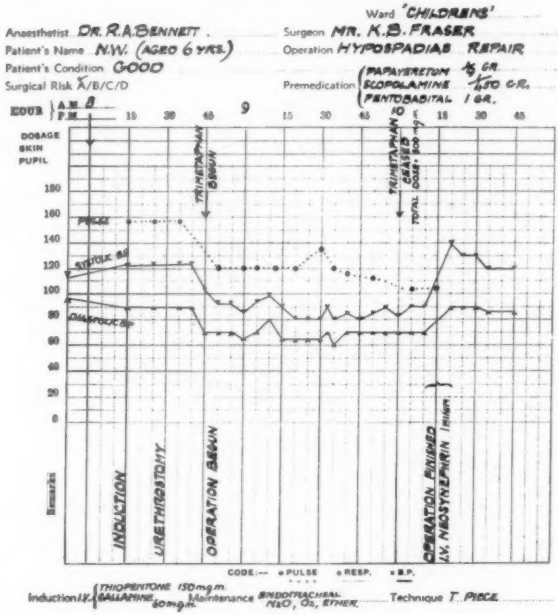
Posturing is most important to ensure a dry operative field. The patient is placed in the lithotomy position. The pelvis is raised on a small rubber pad or cushion, and the table tilted head-down just beyond the horizontal plane.

Five hundred mgm. trimetaphan is mixed with 40 ml. distilled water in a syringe. Intermittent intravenous injections are given to lower the blood pressure and maintain it at a level where bleeding is minimal. This may occur in some cases when systolic pressure in the arm is reduced to 100-105 mm. of mercury, but lower levels are necessary in others and satisfactory conditions may not result until systolic pressures of 80-90 mm. of mercury are produced.

TABLE 2

A TYPICAL ANAESTHETIC CHART FOR THE FIRST STAGE HYPOSPADIAS REPAIR

Anaesthesia Record



Total dose of trimetaphan is usually from 300-350 mgm., the blood pressure being allowed to rise at the end of the operation until normal levels are reached. Occasionally an injection of a vasopressor drug, such as neosynephrine, is required (Table 2).

was removed; in two cases closure was delayed but in each the perineal wound closed slowly and progressively.

In one case the Malecot catheter, after insertion into the bladder, failed to drain; it

TABLE 3

STATISTICAL DETAILS OF THE HOSPITALIZATION

Patient	*Age*	*Dates of Operations*		*Interval between operations*	*Stay in Hospital*		*Total stay in Hospital for the two stages*
		First Stage	*Second Stage*		*First Stage*	*Second Stage*	
I.T.	9 years	July 1956	Oct. 1956	3 months	15 days	8 days	23 days
K.F.	8 ,,	Aug. 1956	Oct. 1956	2½ ,,	13 ,,	8 ,,	21 ,,
R.W.	10 ,,	Oct. 1956	Feb. 1957	4 ,,	12 ,,	8 ,,	20 ,,
K.C.	6½ ,,	Dec. 1956	Mch. 1957	3½ ,,	12 ,,	8 ,,	20 ,,
R.H.	4½ ,,	Dec. 1956	Mch. 1957	3 ,,	12 ,,	8 ,,	20 ,,
S.McL.	10 ,,	Mch. 1957	June 1957	3 ,,	13 ,,	9 ,,	22 ,,
K.R.	5½ ,,	Mch. 1957	July 1957	3½ ,,	10 ,,	10 ,,	20 ,,
N.W.	6 ,,	May 1957	Sep. 1957	3½ ,,	11 ,,	10 ,,	21 ,,
A.S.	6½ ,,	May 1957	Sep. 1957	4 ,,	16 ,,	8 ,,	24 ,,
Average: —	7½ years			3½ months	12½ days	8½ days	21 days

ANALYSIS OF RESULTS OF THE OPERATION FOR HYPOSPADIAS REPAIR

In the nine cases under review the average time in hospital for the first operation was twelve and one-half days and for the second operation eight and one-half days giving a total hospitalization time of twenty-one days (Table 3). This compares favourably with the overall time required for any other method of hypospadias repair.

No serious post-operative swelling was experienced neither was there a serious breakdown of the wound in any case. No case developed a persisting post-operative fistula.

On the two occasions that there was a post-operative urinary leak following the first operation it gave no inconvenience and closed spontaneously after a short period.

No secondary operation of any kind was required once the penis had been satisfactorily straightened and the abnormally placed urethral orifice had been made wide enough for the definitive hypospadias repair to be undertaken.

In most instances the perineal urethrostomy wound closed a few days after the catheter was removed in the first 24 hours and the urine was by-passed satisfactorily through the urethrostomy wound (Table 4). It was not found necessary to pass sounds in any case after operation.

The post-operative urinary stream in every case is full and strong without any spraying and there has been no suggestion in any case, of diminution in the force and volume of the urinary stream with the passage of time (Fig. IX).

Advantages of the operation

1. The likelihood of persisting post-operative fistula, the constant enemy of every surgeon undertaking this work, is minimized because, after the first stage of the operation has been completed, the new urethra lies buried deeply in the scrotal tissues far away from any sutures communicating with the surface; this means that subsequently any urine which may escape from the newly formed urethra has to traverse a considerable distance to reach the surface; as a consequence, any such leak ceases spontaneously before an epithelialized track can establish itself as a permanent fistula.

TABLE 4

THE POST-OPERATIVE COURSE OF EACH CASE IN THE SERIES

Patient	*Complications*	*Persisting Post-Operative Fistulae*
I.T.	Nil	None
K.F.	Nil	None
R.W.	Nil	None
K.C.	After the first operation a small urinary leak developed on the left peno-scrotal suture line; the leak closed spontaneously in fourteen days.	None
R.H.	Nil	None
S.McL.	Slow closure of perineal urethrostomy wound: an infected haematoma under the distal end of the flaps in the second stage slightly delayed final healing but not hospitalization time.	None
K.R.	Nil	None
N.W.	A small superficial slough slightly delayed healing in the second stage.	None
A.S.	(A blind boy — very hard to nurse.) The Malecot catheter obstructed and was removed on the first post-operative day; the urethrostomy wound continued to by-pass the urine successfully. After the first operation a urinary leak occurred on the left peno-scrotal suture line which closed spontaneously in twenty-one days. Perineal urethrostomy wound slow to close.	None

This desirable feature of having the new urethra separated by as great a distance as possible from any surface suture line was mentioned by Denis Browne in 1936 before he commenced to use his present technique.

2. The new urethral lumen is satisfactorily large and does not need any subsequent dilation.

3. The new urethra consists of skin in continuity, and an undesirable suture line between the old and the new urethra is thus obviated.

4. The new urethra is completely lined with epithelium *ab initio*, a fact which effectively minimizes any fibroblastic reaction.

5. There is no likelihood of hair formation in the new urethral tube, for the skin used in its formation has come originally from the prepuce and so is free of hair follicles.

6. The new urethra itself and the new ventral skin surface of the penis are composed of highly elastic tissue which adapts itself very readily to erection without producing any penile distortion.

7. The final result gives a penis which is not only straight but one which has good ventral length and a well-marked peno-scrotal angle.

8. There is very litle oedema following either stage of the hypospadias repair, partly because of the limited amount of undermining of flaps required and in the first stage largely as a result of the hypotensive anaesthetic.

9. Nursing is easy and the patients are remarkably free from discomfort.

10. The overall time in hospital is satisfactorily short.

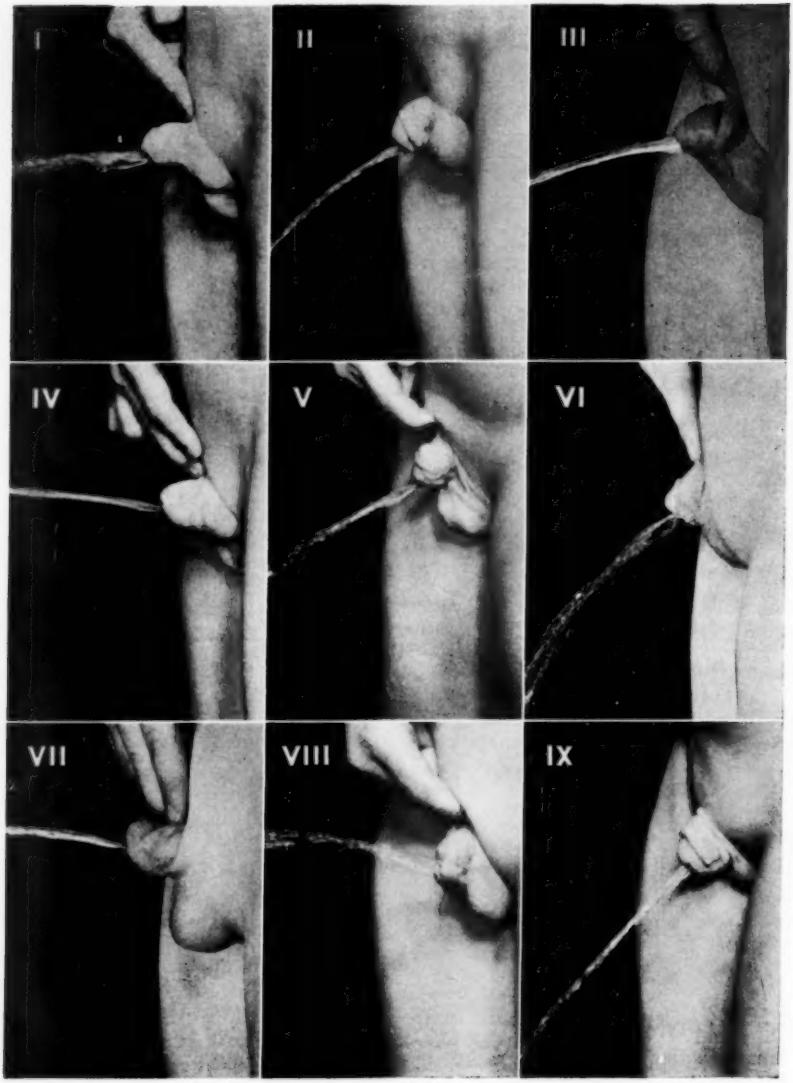


FIG. IX. The nine boys, each photographed some months after the completion of his operation, showing the fullness and forward direction of the urinary stream and the absence of spraying in every case.

Patients: (i) I.T., (ii) K.F., (iii) R.W., (iv) K.C., (v) R.H., (vi) S.McL., (vii) K.R., (viii) N.W., (ix) A.S.

Disadvantages of the operation

1. The new urethral orifice is not quite in the position of the normal orifice. This is of no functional import and eliminates the possible danger of haemorrhage from burrowing through the glans penis. It may be argued that this is a slight cosmetic disadvantage but actually it is hard to satisfy oneself that these patients are not passing urine from a perfectly normally placed urethra.

2. Two operations are needed for the hypospadias repair; this is unimportant, for the overall hospitalization time is only three weeks and the discomfort at any stage is minimal.

3. The operation requires a scrotum of adequate size and is not suitable for cases with double undescended testes, until such time as these have been brought into the scrotum.

DISCUSSION

It is not suggested for a moment that the method here described is the only one that will give satisfactory results in hypospadias repair. It is well known for instance, that many surgeons have successfully employed inlay grafts with subsequent closure of the fistula between the old and new urethra, a method first described by Nove Josserand in 1897 and subsequently modified by McIndoe (1937, 1948), Havens and Black (1949) and Gross (1953). It cannot be denied however, that any method using inlay grafts involves the use of skin obtained from an area remote from the penis and unlike it in texture. McIndoe (1937) while pointing out the merits of Thiersch grafts from the inner side of the arm, admits that the non-extensibility of such a free graft used to fashion the new urethra is a definite disadvantage. Vilray Blair *et alii* (1933) stressed the point that no tissue makes a satisfactory restoration of the urethra except that which comes from the penis or scrotum. Undoubtedly there often seems to be some anxiety about contracture in the early post-operative period when using this method and these authors all employ an obturator in the new urethra continuously for six months after operation; this must surely be an irritating necessity from the patient's point of view. Another disadvantage is that this operation is often delayed until puberty or even later, in order to make more certain of satisfactory results.

The claims that severe psychological trauma and even psychopathic deviations occur as a result of delaying operation until after puberty, have probably been exaggerated. It cannot be denied however, that, given the choice, any boy suffering from hypospadias would much prefer to have his surgery behind him early, so that he could face his school days free from any physical disability. Edmunds (1926) put the position succinctly in his Hunterian Lecture when he said: "It requires little imagination and knowledge of the average small boy to realize that a schoolboy with hypospadias would be seriously handicapped and would be liable to unkind comments from his schoolmates."

By adopting the method described here, which can be completed at or very shortly after the time the boy reaches school age, he is certainly saved many years of self-consciousness and embarrassment.

Young and Benjamin (1949) have reported their results in 16 cases operated on in the pre-school age using inlay grafts, eliminating the continued use of an obturator after the repair, and joining the new urethra to the old at the same operation. Eleven of these cases developed a fistula; a fistula was still present in 3 out of 7 of these after secondary closure had been attempted. On these results the method does not appeal as a routine procedure. Davis (1940) described the use of a tube-pedicle from the dorsal skin of the penis—based proximally—in order to fashion a distal urethra through the glans. Having done this as an initial step he then proceeded to do what was virtually a Cabot repair. The method is ingenious but the technical difficulties and dangers involved for what is a very doubtful gain do not warrant its adoption.

SUMMARY

What I have attempted here is to present a technique for hypospadias repair which, carried out in 9 consecutive cases over a period of fourteen months, has been subjected to critical analysis.

The results functionally, and cosmetically have been satisfactory. The longest overall stay in hospital was twenty-four days, no serious nursing difficulties have been encountered and the patients have all been remarkably comfortable in the early post-operative period.

The most pleasing features have been firstly, the sureness of primary healing with complete absence of any fistula formation or of any complication requiring further surgery; secondly, the ease of nursing; thirdly, the short overall period of hospitalization; fourthly, the early age at which the repair

can be completed and finally, the construction, in every case, of a penis which is functionally perfect and anatomically almost indistinguishable from normal.

ACKNOWLEDGEMENTS

Thanks are extended to Dr. Roger Bennett who was responsible for the anaesthetics, and has written the notes on the hypotensive technique; to Miss Lyndsey Pegus, Medical Artist at the University of Queensland Medical School, who did the drawings and tables; and to the Photographic Department of the University of Queensland.

Finally, it is desired to pay tribute to the many surgeons, past and present, in various parts of the world, some of whom are mentioned here but many of whom remain nameless, whose pioneer work has advanced the surgery of hypospadias repair to such a degree that, with care and perseverance, a very high percentage of initially successful results can be attained.

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NON-PARASITIC CYSTS OF THE LIVER

By Reginald Webster

Royal Children's Hospital, Melbourne

In Australia the discovery in the liver of a solitary cyst of appropriate dimensions naturally suggests hydatid disease, and such presumptive diagnosis must remain in the forefront of probability until the nature of the cyst is elucidated as other than parasitic. Non-parasitic hepatic cysts, however, occasionally emerge as productive of usually vague, but on rare occasions dramatic, abdominal symptoms and signs, though many remain innocuous, their presence unsuspected and ultimately revealed as incidental findings in the course of routine autopsy work.

For this reason, as well as the oblivion which may be presumed to have been the fate of many such cysts as the result of failure on the part of clinicians to record them, the number of reported instances is comparatively few, and without doubt understates the frequency of occurrence of non-parasitic hepatic cysts. Caravati *et alii* (1950) state that no more than 200 cases of solitary liver cysts other than parasitic have been recorded. It has been established that the cysts may be disclosed at any time during life, from the pre-natal period up to and beyond the traditionally allotted span of three score years and ten. It is a curious fact, attested by many writers, that the cysts exhibit a decided predilection for the female sex, the agreed ratio of females to males being 4 : 1. Because of their clinical silence hepatic cysts of the type under consideration obtrude themselves relatively seldom in childhood. Montgomery (1940) could find reports of only 27 examples of solitary non-parasitic cysts of the liver as having occurred in patients under the age of 13 years, prior to his report of two additional cases in childhood. Munroe (1942) published details of a non-parasitic cyst of the liver as he studied it in an infant aged 9 months, and another such cyst, which measured 12 x 20 cm. in maximal dimensions was reported by Desser and Smith (1956) as having occurred in the liver of a baby admitted to the Los Angeles Children's Hospital. This experience led Desser and Smith to institute a search of the records of the hospital with the result that there were discovered case histories of six other children in whom the presence of non-parasitic hepatic cysts had been recorded.

The two examples of solitary non-parasitic cysts of the liver which it is proposed to discuss have occupied their places in the museum of pathology at the Royal Children's Hospital, Melbourne, since 1943 and they are the only two specimens of their type that I have been enabled to impound for the museum in a period of forty years' association with the Department of Pathology of the Royal Children's Hospital. By a curious chance the specimens were obtained within a few months of each other, but it cannot be safely concluded that only two hepatic cysts other than hydatid have occurred during this long period; cysts considered irremovable and successfully treated by marsupialization would have passed into the records leaving no concrete memorial.

Specimen 1

The first example of a non-parasitic cyst of the liver to find its way to the pathology department was provided by a boy aged 9 years. Six months prior to his admission to the Royal Children's Hospital his parents had sought the advice of their family doctor regarding the vague adbominal discomfort and "biliousness" of which the lad complained. The practitioner consulted drew attention to a swelling in the epigastrium, but as the symptoms seemed to improve no further action was taken in the ensuing six months.

The boy first attended the Royal Children's Hospital on 4th June, 1943, when on physical examina- it was found that underlying a certain fullness in the epigastrium was a palpable rounded "tumour," which could not be defined from the margin of the left lobe of the liver. The most probable clinical diagnosis was that of hydatid disease, but this was not supported by the results attending the Casoni intra-dermal test and the complement fixation reaction. Exploratory laparotomy undertaken by Mr. Eric Price revealed a large cyst, intimately related to and apparently arising from the inferior surface of the left lobe of the liver.

After aspiration of a large quantity of its fluid content it became apparent that the cyst was bilocular, and that it occupied virtually the whole of

the left lobe of the liver, and a small segment of the right lobe. Although in its major portion intra-hepatic the cyst was successfully removed, and with the exception of an outlet for the passage of a drainage tube, the cavernous cyst bed was closed by the apposition of its margins with catgut sutures.

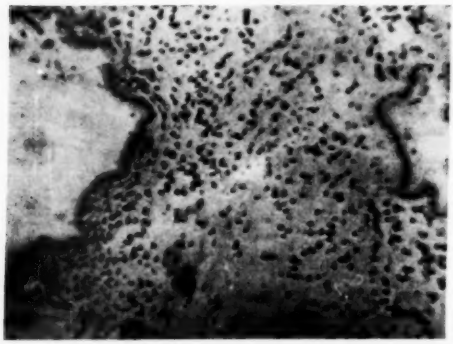

FIG. I. Cystic spaces within the wall of a non-parasitic cyst of the liver; lining of cuboidal epithelium.

In the laboratory examination of the fluid withdrawn from the cyst, a search directed towards the detection of scolices, hooklets, or laminated membrane, the finding of any of which would have infallibly identified the cyst as an hydatid cyst, was barren of result. The fluid was turbid, gave no impression of the presence of bile, and in the microscopic examination of the centrifuged deposit exhibited a few intact epithelial cells, some cholesterol crystals, and much amorphous debris. In the partially collapsed condition in which it reached the pathology department the cyst was roughly spherical in shape, and measured 9 centimetres in diameter. Small subsidiary cysts were noted here and there in the otherwise stout wall, and a thick fibrous septum divided the cavity into two main loculi.

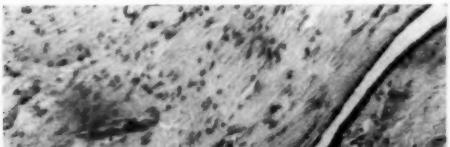

FIG. II. One of the many epithelium-lined intramural clefts.

The examination of microscopic sections of the wall of the cyst showed that the intima was devoid of epithelial covering, and was more cellular than the dense hyalinized fibrous tissue which composed the major portion of the wall. Within this zone were to be found at frequent intervals clefts and spaces of bizarre shapes and varying sizes, all lined by a low columnar or cuboidal epithelium, sometimes flattened and occasionally detached, but on the whole of clearly defined and well sustained morphology (Figs. I and II). The character of the epithelium, and the rare appearance among the intramural cysts of a channel indistinguishable with respect to the size of its lumen and the quality of its epithelium from a normal bile duct suggested very strongly that the cyst was the ultimate expression of a vagary in the development of the biliary ducts.

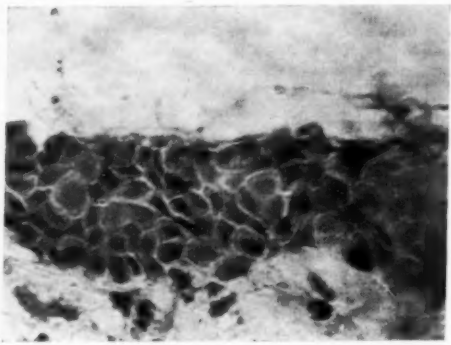

FIG. III. Squamous metaplasia affecting the otherwise consistently cuboidal epithelium lining the major and minor loculi.

In Fig. III the otherwise universal single layer of cuboidal epithelium is seen to have undergone squamous metaplasia, an observation to which no significance, other than that the occurrence illustrates yet again the effect of abnormal conditions in evoking a latent potency in well differentiated epithelium, is to be attached.

It remains to add that the post-operative course in the boy from whom this cyst was removed was uncomplicated and he was discharged from hospital on 7th July, 1943, twenty-five days after his admission.

Specimen 2

The second specimen of hepatic cyst was secured at autopsy on the body of a girl, aged four years, who had first become ill late in the month of March, 1943. She was admitted to a provincial base hospital on 13th April, jaundiced and febrile, having been ill for two weeks. Prompt defervescence and concurrent fading of the icterus seemed to warrant a diagnosis of "catarrhal jaundice," and the little girl was discharged from hospital at the end of one week.

She was re-admitted to the country hospital on 28th June on account of recurrent jaundice of three weeks duration. Her temperature at that time was 104°F (40°C), and the total leucocyte count 36,000 per c.mm. The Van den Bergh test applied to the blood serum resulted in the "immediate direct" type of reaction. Laparotomy was performed on 8th July when it was observed that the gall-bladder, though full, could be emptied readily by pressure. Aspirated bile yielded no growth on attempted cultivation, and thereafter apparently normal bile, as much as 180 cc. per day, drained from a cholecystostomy opening.

The child was referred to the Royal Children's Hospital on 10th August by the late Dr. H. Douglas Stephens. She was then very ill; the number of leucocytes reached the high figure of 76,900 per c.mm., and a deep degree of jaundice and sustained high fever were conspicuous clinical features. Serological and radiological investigations were directed towards confirmation of a suspected hydatid cyst as the basis of the jaundice indicated clearly as obstructive by the clay coloured stools. Negative findings were returned for both the complement fixation test and the Casoni intra-dermal reaction. A report dated 10th August, 1943, recorded that the chest was radiographically normal, that the diaphragmatic dome was normal in position and outline, but that a small ovoid density overlay the lateral aspect of the right lobe of the liver. A footnote to the report requested an opportunity to repeat the postero-anterior radiograph of the liver, and to secure also a lateral view of the right lobe and the right dome of the diaphragm. In a revision of the radiographic report issued on 17th August, it was stated that the shadow previously noted in the upper segment of the right side of the abdomen was no longer in evidence; it was dismissed as an artefact and a final statement issued that the radiographic examinations disclosed no evidence of hepatic (or pulmonary) hydatid.

During the six weks that the patient remained in the Royal Children's Hospital the biliary fistula closed. Dr. Stephens contemplated further laparotomy, which complicating pneumonia and the extremity of the child's illness obliged him to defer. She died on 20th September, emaciated, cachectic, and deeply jaundiced.

At autopsy the liver was seen to be greatly enlarged and of the deep olive-green colour induced by prolonged biliary obstruction. The gall-bladder contained mucoid material in which no bile pigment was apparent. Emerging from the inferior surface of the right lobe of the liver, and so situated as to impinge upon and stretch the free edge of the gastro-hepatic omentum, was a tense cyst, the presence and location of which provided an abundantly adequate explanation of the obstructive jaundice. The cyst was found to arise within the substance of the liver, though it was not very deeply imbedded. Contrary to the impression gained by first inspection the cyst was multilocular; its fluid content was frankly purulent.

Throughout the liver were to be seen multiple abscesses and channels of suppuration; bile-stained pus was expressible from the hepatic ducts, and the parenchyma of the liver was soggy and friable throughout. The pathological condition in the liver was obviously that of widely distributed suppurative cholangitis.

As has been indicated the offending cyst was multilocular, and approximately one-fourth of it was imbedded in the hepatic parenchyma; its dimensions after the dissection necessary to excise it were 12 x 7.5 cm. In transverse section it presented two main chambers surrounded by a cluster of minor loculi. Microscopic sections of the cyst showed the several loculi to be lined by cuboidal epithelium; dispersed through the intervening fibrous septa and the outer limiting wall of the cyst, and lined with epithelium of consistently cuboidal morphology, were an astonishing number of lacunae, exhibiting great diversity of size and every conceivable modification of shape between wide open spaces and mere slits or fissures. The epithelium throughout was surprisingly well preserved although that of the large central loculi had suffered detachment and a degree of destruction as the result of infection and concomitant rise in intra-cystic tension.

PATHOGENESIS OF NON-PARASITIC CYSTS OF THE LIVER

It has been customary to differentiate sharply between single or "solitary" hepatic cysts such as those which form the basis of this communication, and those which by their multiplicity constitute generalized cystic disease of the liver, an age-old entity commonly associated with polycystic kidneys, and less frequently with cysts of the pancreas, lungs, spleen, and brain; malformations which have been noted as linked with cystic disease of the liver include hare-lip, cleft palate, meningocele, and hyperdactylism. None of these fellow travellers has been identified with "solitary" hepatic cysts and majority opinion in the past has been that lonesome "simple" cysts of the liver have nothing in common with those of smaller size which honeycomb the liver in general cystic disease.

In an early communication on non-parasitic cysts of the liver, Boyd (1913) proposed and favoured the view that universal cystic disease and solitary cysts of the liver were different manifestations of the same disease, or in other words, were respectively the outcomes of a process the incidence of which might be diffuse or focal. The essential identity of the two manifestations of cystic disease of the liver from the point of view of pathogenesis is suggested by reports,

noted by Stoesser and Wangensteen (1929), of the occurrence of small cysts in the proximity of one of dominating size, and at first sight solitary. Sonntag (1913), whose classification of hepatic cysts has been widely quoted, considered favourably the suggestion advanced by Boyd. Until the question is resolved, however, the contrasts existing between "solitary" cysts and general cystic disease of the liver, particularly as regard amenability to treatment and association with cysts in other organs and diverse malformations, render it advisable to obviate confusion by treating focal and diffuse cystic disease of the liver as separate clinical entities.

The epithelial lining of the two cysts described in this communication, a lining which sustains its character throughout their manifold intra-mural clefts and spaces, suffices to exclude both specimens from the categories of teratomatous cysts, cavernous lymphangiomatoid structures, and the pseudo-cysts which result from degenerative processes. Further, the character of the epithelium, and the occasional appearance in the microscopic sections prepared from Specimen I, of channels of the calibre and epithelial lining of normal bile ducts, render irresistibly strong the presumption that the cysts are the ultimate expressions of deviation from the normal evolution of the biliary system.

Current concepts of the histogenesis of non-parasitic cysts of the liver all postulate a developmental defect affecting the bile ducts, by which these channels are anatomically aberrant, either in the sense that some are extra-hepatic, or that certain bile ducts within the liver traverse the hepatic parenchyma independently of the portal tracts, and fail to establish communication with the biliary canaliculi running between the cords of cells of which the liver lobule is composed.

Since the publication of his closely reasoned thesis, Moschcowitz (1906) has figured as the protagonist of the view that hepatic cysts are derived from aberrant bile ducts, either extra- or intra-hepatic. As authenticating the occurrence of extra-hepatic bile ducts, Moschcowitz adduced chapter and verse from Quain's textbook of anatomy, in which classic treatise he found the statement that extra-hepatic bile ducts were first described by Ferrein and Kiernan as occasionally occurring "in the left lateral ligament and in the fibrous cords bridging the fossa for the vena cava, and the fissure for the umbilical vein. They anastomose together in the form of a network, and are accompanied by branches of the vena porta, hepatic artery, and hepatic vein." Moschcowitz also cited Renault as having found wayward bile ducts on the under surface of the liver, and named ten authors who had described smaller or larger cysts containing clear watery fluid, on the inferior aspect of the surface of the liver. Many showed a trabecular formation within the cyst, others a smooth lining, but bile ducts were found in the connective tissue walls of the cysts, and in the pedicles by which the extra-hepatic cysts were attached to the capsule of the liver.

It requires a consideration of the embryology of the liver to formulate any hypothesis regarding the histogenesis of intra-hepatic bile ducts which, in that they are not integrated with the normal biliary duct system, and fail to establish communication with the biliary canaliculi, may properly be termed aberrant. It is necessary to recall the first appearance of the liver as a diverticulum which arises ventrally from the endoderm of that portion of the gut destined to form the duodenum. A maze of branching and anastomosing cell cords grows out from the diverticulum, the distal portions of the cords developing in due course as the secretory tubules of the liver, and their proximal portions as the hepatic ducts. The young hepatic tubules grow between the layers of the splanchnic mesoderm, which invest the developing liver and give rise to its fibrous tissue capsule and all the supporting tissue of the liver lobules, as well as the connective tissue and smooth muscle layers of the system of ducts.

The branching and anastomosing tubules ultimately productive of the actively secreting parenchyma of the liver, are distal continuations of the hepatic ducts, and the pattern of the branching of the growing hepatic tubules is quite characteristic (Patten, 1953). From each of the primary cell cords a series of branches grows out at a right angle. Each

of these branches in turn sprouts a system of radiating smaller branches, which become the tubules of a secretory lobule of the liver. The axial cord of cells from which they arise serves as a branch of the hepatic duct system, which drains such a lobule into one of the main ducts leading to the gall-bladder.

If as the result of the operation of any or all of the obscure factors which impede or modify the normal course of development, the proliferating endodermal cells of the distal portions of the hepatic cords should fail to differentiate into hepatic parenchymal cells, while development in the axial cords proceeds normally to elaborate the epithelium lined channels of the bile ducts, there would be brought about a state of affairs in which unorientated and non-communicating biliary ducts would be found occupying mesoderm which normally should have been replaced by endodermal cells differentiating ultimately into the columns of secreting cells which distinguish the hepatic lobule; from which hypothesis the suggestion emerges that the fundamental defect in cystic disease of the liver is hypoplasia involving the parenchyma, a deficiency in which the system of hepatic ducts does not participate. Such a developmental anomaly may conceivably be focal in its incidence, or operate over fields of varying extent, possibly limited only by the confines of the embryonic liver.

In this connexion I have been enabled, by the courtesy of Mr. F. Douglas Stephens, to repeat an observation of Moschcowitz in the examination of microscopic sections of the liver of a stillborn baby who exhibited polycystic disease (or malformation) affecting both kidneys, and a hypoplastic and cystic state of the pancreas. In view of the well-known association of generalized cystic disease of the liver with polycystic kidneys, and particularly as the liver displayed no macroscopically appreciable departure from the normal, microscopic study of this viscus held promise of a clue to the solution of the problem of cystic disease of the liver by uncovering the process in an incipient phase. The histology revealed in microscopic sections of the liver was indeed remarkable. The liver cells were normal, but traversing the parenchyma in every direction were countless well-formed biliary ducts, both within and without the capsules of Glisson. The ubiquitous biliary ducts were separated from the surrounding liver trabeculae by a circular investment of fibrillar connective tissue, and throughout all sections fibrous tissue was in excess (Fig. IV).

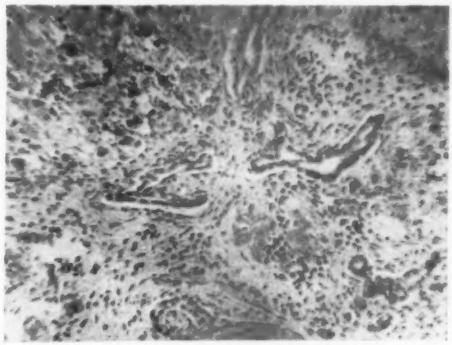

FIG. IV. Photomicrograph of the liver of a stillborn infant who exhibited polycystic kidneys (see text).

In short, the microscopic sections, with their multiplicity of bile ducts and general fibrosis closely approximated those of postnatal acquired biliary cirrhosis; they could fairly be described as depicting congenital biliary cirrhosis, with the reservation that the fibrosis was not the result of an infective process, but represented the natural evolution of mesoderm, the persistence of which was due to the failure of its occupation to a normal degree by endodermal cells developing as hepatic parenchyma. It can well be imagined that many biliary ducts participate in the general dysplasia to the extent that they are obstructed at various points as the result of imperfect canalization, and thus Moschcowitz, having considered and rejected any process of neoplasia such as cystadenoma, reached his conclusion that the cysts of the polycystic liver, so commonly associated with kidneys and pancreas in similar state, were retention cysts of aberrant bile ducts. Confirmation of this view is provided by Fig. V, which reproduces a photomicrograph of a field chosen from the same microscopic section as supplied Fig. IV. In a channel lined with cuboidal epithelium and bathed in peri-canalicular oedema, the process of biliary duct dilatation is seen to have been initiated.

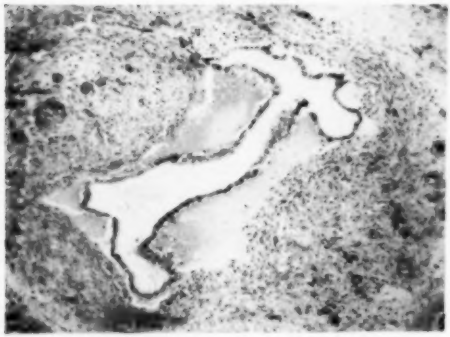

FIG. V. Another field from the microscopic section which provided Fig. IV, showing initiation of cystic dilatation of a biliary duct.

It seems plausible, and indeed reasonable to assume that the process of liver cell hypoplasia, with the persistence of a normal number of biliary ducts of necessity not integrated with secreting lobules, developmentally obstructed and prone to become cystic, may be focal or diffuse in its incidence. If focal there may be expected to ensue a "solitary" cyst, possibly attended by one or two small satellites, and if diffuse, more or less general cystic change in the liver would be indicated as the natural outcome. The conception advanced of the histogenesis of hepatic cysts, solitary and multiple, lonesome and gregarious, is in line with the suggestions of Boyd and of Sonntag, that the so-called "solitary" non-parasitic cysts of the liver, and widespread cyst formation in this viscus represent different degress of one fundamental process.

CYSTADENOMA OR HAMARTOMA?

In the literature relating to the "solitary" cysts are to be found many authoritative expressions of opinion that such cysts are neoplastic in nature and represent cystadenomata of the bile ducts; they have been held to be analogues of the ovarian cystadenoma. Siegmund and Borrmann are cited by Moschcowitz (1906) as propounding this view, not only with regard to solitary cysts but as applicable also to those which riddle the liver in general cystic disease. Borrmann postulated a disturbance in the relative rapidity of growth of the epithelium and connective tissue of the bile ducts, by which the growth of the former outstripped that of the latter; at the same time he assigned an active role to the connective tissue, regarding it as an integral component of a fibro-adenoma. He therefore described liver cysts as fibro-adenomata, and placed them in the same category as the corresponding tumours of the breast and ovary. There have been many doughty protagonists of the view that a neoplastic process underlies the development of hepatic cysts. Stoessen and Wangensteen (1929) quote Kaufman, Konjetzny, Sternberg and others as designating the condition "cystadenoma hepates." Ewing (1928) considered that a congenital malformation was the original factor, and that on the basis of a developmental anomaly progressive dilatation of the implicated bile ducts might occur or adenomatous processes be established.

Undoubtedly true cystadenomata of the bile ducts may arise, but they are rare tumours. The description by Keen (1892) of a tumour which he removed in 1891 from the liver of a woman aged 31 years, would seem to leave no doubt of its nature as a true multi-locular cystadenoma. It weighed 113 grammes and contained many cavities lined by cylindrical cells and supported by fibromuscular tissue. In Siegmund's patient an isolated cystic mass in which were many microscopic fields suggesting active neoplastic growth of cubical cells, formed a large part of the liver. But such tumours, which may be allowed as true cystadenomata of the bile ducts, occur very infrequently, and have little in common with the entity of which the two specimens described in this paper are presented as examples.

In neither of the two "solitary" cysts of the liver, selected from the museum of pathology of the Royal Children's Hospital for description and discussion, of identical and relatively simple histology, and essentially multi-locular, has there appeared any microscopic evidence of the neoplastic process connoted by the term cystadenoma. In a search through serial microscopic sections prepared from a block of tissue 2 cm. in thickness and embracing a complete transverse section of the obviously multi-locular cyst No. II, I have been unable to find any outward sprouts from the epithelium lining

the cavities which abound in the wall of the cyst; nor could I observe more than one layer of epithelium in the major cyst cavities, or in any of the intra-mural cysts, much less papilliform epithelial processes projecting into their lumina. If the suggesion of a developmental defect as the basis of hepatic cysts, attributable originally to Still (1898), and elaborated in earlier paragraphs of this paper be recalled, it will be appreciated that liver cell hypoplasia and persistence of bile ducts constitute that abnormal blending of tissues, with excess of a particular component, which is the very essence of a hamartoma. And it is as hamartomata that I present the two specimens on which I have ventured to write these notes, which concern primarily so-called "simple" solitary cysts of the liver, distinguished by the presence of a multiplicity of distorted bile ducts in their fibrous tissue walls. If the proposition be accepted that solitary hepatic cysts and those of generalized cystic disease of the liver represent the extremes of the manifestations of one fundamental process, it follows that the polycystic liver is to be regarded as a congeries of hamartomata, a conception in which I see nothing incongruous or illogical.

In connexion with the neoplastic nature of hepatic cysts, presumed by those who would designate them biliary duct cystadenomata, Munroe (1942) makes the pertinent observation that if the tortuous bile ducts deep in the walls of such cysts were truly blastomatous in nature, many cysts might have been expected to show recurrence after the marsupialization enforced by insuperable difficulty attending their eradication. Such has not been the history of the surgery of "simple" non-parasitic cysts of the liver.

General Considerations Concerning "Solitary" Cysts

As already indicated here is no particular age group in which a "solitary" hepatic cyst may be expected to declare its presence, nor does there seem to be any limit to the size which they may occasionally attain. They may grow to great size, even in the foetus, and obstruction to delivery by a large cyst of the liver has been recorded. Their predilection for the female sex has been noted. They have been found most frequently at the antero-inferior margin of the right lobe of the liver, occasionally attached to the round ligament. Exceptionally a solitary cyst of the liver is pedunculated, in which event removal is facilitated but a risk of torsion introduced. There is as a rule, little or no clinical evidence of the presence of a simple hepatic cyst, a fact which has been attributed to the relatively low intracystic tension which permits neighbouring viscera to accommodate themselves to its presence and evade pressure effects during a long period of slow enlargement of the cyst. Acute symptoms may follow suppuration or intracystic haemorrhage, traumatic or spontaneous. Kilvington (1902) reported the death of a patient from haemorrhage into a large simple cyst of the liver; the clinical features associated with the catastrophe resembled those attending rupture of a large internal aneurism.

The cyst may lie deeply in the substance of the liver and destroy most of the right lobe, in which event it is not amenable to surgical removal. Although the subject of treatment lies outside the scope of this paper, it may be noted that when removal is not feasible, marsupialization, dissection of the lining, and anastomosis to the stomach have all been successfully attempted. Manheimer (1953) in reporting the successful treatment of a large solitary cyst of the liver, urged that whenever total excision of a liver cyst appears hazardous, internal drainage by anastomosis to the stomach or proximal portion of the duodenum, is an eminently satisfactory procedure. He advocated that marsupialization be superseded by this measure as the accepted method of dealing with irremovable cysts.

Summary

1. The clinical manifestations of two examples of "solitary" non-parasitic cysts of the liver, as they occurred in children aged nine and four years respectively, are described, and the pathogenesis and essential nature of such cysts discussed.

2. The distinguishing feature of the histology of non-parasitic hepatic cysts is the presence within their fibrous tissue walls of numerous subsidiary cysts lined by epithelium strictly comparable with that of biliary ducts.

3. It is affirmed that non-parasitic hepatic cysts are referable to a developmental anomaly, and consideration is given to the nature of the deviation from the normal development of the liver and its system of biliary ducts.
4. That "solitary" cysts and polycystic disease of the liver represent different degrees of the same process of maldevelopment is advocated as a rational conception.
5. It is concluded that with few exceptions non-parasitic cysts of the liver are more correctly regarded as hamartomata than as biliary duct cystadenomata.

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MEASUREMENT OF UPPER LIMB VOLUMES: A CLINICAL METHOD*

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Introduction

SIGNIFICANT swelling of the upper limb may occur in a wide variety of pathological conditions including infections, obstruction of the venous or lymphatic channels by new growths, in giant urticaria, and following extensive removal of the efferent lymphatics as in radical mastectomy with axillary node dissection. For a detailed study of the clinical pathology it may be desirable to have a quantitative record of the degree of this swelling. Also serial readings may be of value in following the natural history of the swelling, the factors affecting it, and the efficacy of various methods of treatment.

The object of this paper is to describe a method used in this laboratory for measuring upper limb volume. The volume of the limb and its segments in normal adult women and in patients with post-mastectomy lymphoedema has been examined but in this paper only data from the former group are given in detail. The method described has proved to be simple and reliable in practice and examples of its application are given.

Apparatus and Method

The apparatus is based upon the simple physical principle illustrated in Fig. I.† The upper limb is immersed in water in the tank, A, filled just to overflowing. As the hand and limb are immersed, water immediately escapes down the overflow spout, OS, into a 2,000 ml. graduated collecting cylinder, B. The volume of water displaced into the collecting cylinder is a measure of the volume of the limb to the depth immersed.

Fig. I is a sectional view of the apparatus illustrating its main features. The tank is cylindrical and is constructed of $\frac{1}{4}''$ Perspex cemented to a square Perspex base, $\frac{1}{2}''$ thick, fitted with levelling screws, LS, at each corner. Near and parallel to adjacent edges of the base are small spirit levels, the screws and spirit levels being used to maintain the level of the tank constant from day to day. The internal diameter of the tank is 8″ and its height 2′ 6″.

The small ledge, F, is a horizontal shelf on which the patient's middle finger rests firmly as the limb is immersed, the palmar surface of the hand and volar surface of the limb being held against the metal rule, R, which slides freely up and down in a housing, RSG, attached to the wall of the tank. The metal rule is T-shaped in section. The horizontal limb of the T faces into the tank and is marked off in inches, the vertical limb of the T being snugly gripped in the "travel" of the rule support guide, RSG.

At the top edge of the tank the housing is fitted with a hand wheel, HW, which, when screwed tight, fixes the rule (and the ledge F) at any desired depth. On the cursor block, CB, also incorporated in the upper end of the housing, is engraved a horizontal line which runs to the edge of the rule. When the upper surface of the finger rest is in the same horizontal plan as the lowermost edge, C, of the overflow spout (and therefore in the plane of the meniscus, M) the cursor line reads zero on the rule. Thus as the finger rest is immersed in the water its depth of immersion (and that of the upper limb if the outstretched middle finger is kept in contact with F) is read off in inches at the cursor line.

*Work done under a grant-in-aid from the New South Wales State Cancer Council in the Joan Bonamy Surgical Research Laboratory.

†We are indebted to Mr. Peter Lawson for his help with the design and construction of the apparatus.

If, for example, it is desired to record the volume of the hand and forearm, a skin pencil mark is made on the tip of the olecranon. The limb is then immersed as described, pushing the finger rest and rule smoothly down in its housing, until this pencil mark is level with the meniscus. The hand wheel is then tightened and the reading at the cursor line is noted, the value constituting the depth setting for all future forearm volume measurements on the patient concerned. As soon as overflow has ceased the volume of water displaced into the measuring cylinder is read off and recorded.

If the arm volume is also required, the hand wheel is loosened again and the rule and ledge are pushed steadily down by the patient's outstretched middle finger until he cannot reach further into the tank because his axillary folds are bearing on its upper margin. The hand wheel is tightened and the new depth setting is recorded; it is used in all future measurements of the patient's whole upper limb volume. When overflow into the measuring cylinder has ceased the additional volume of water displaced is noted and this gives the arm volume. Hand and forearm plus arm volumes of course give the volume of the whole upper limb.

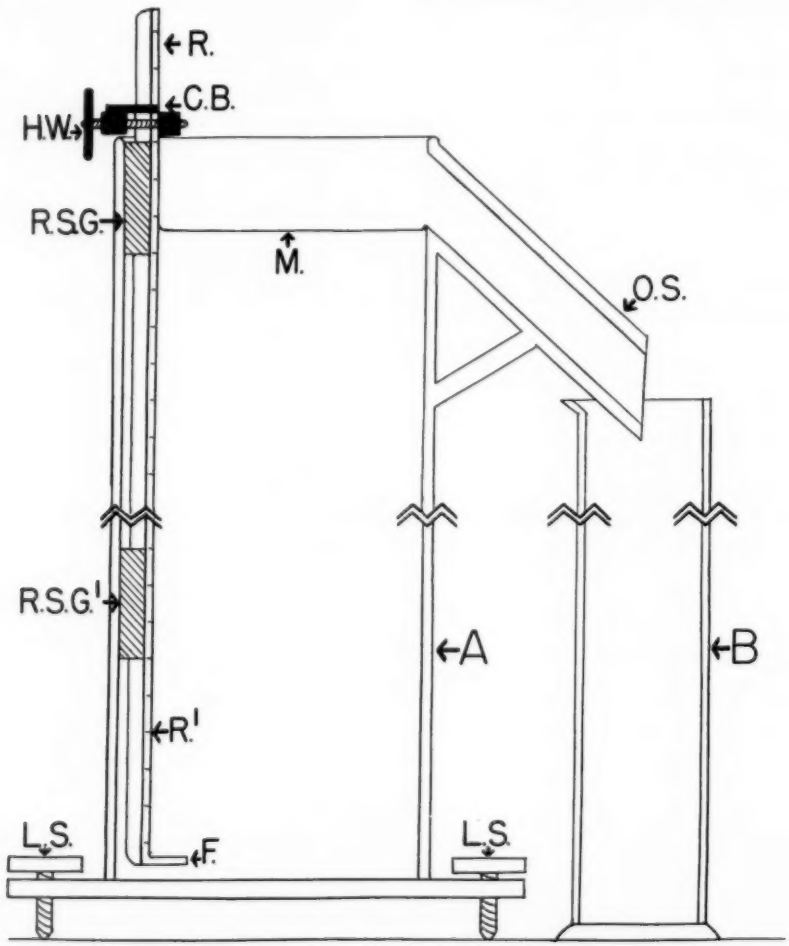


FIG. I. Sectional diagram of apparatus for measurement of limb volume.

In patients with pathological swelling of one upper limb, the volume of the affected limb should first be measured and the depth settings determined therefrom because the maximum depth to which it can be immersed in the tank is often less than that possible on the healthy side where arm and shoulder movements may be freer. For comparison of the volume of the two upper limbs and of their segments, the depth settings used on the two sides must of course be identical.

quantity of detergent is added to the water since this lowers surface tension and reduces errors due to the meniscus. (iii) The tip of the middle finger, and not that of a long finger-nail, should rest against the platform F, otherwise a distinct error will arise especially in whole limb measurements. (iv) As the limb is immersed, the finger, wrist and volar surface of the limb are held in contact with the rule. This ensures a standard position of the limb in the tank, and makes

TABLE 1

REPRODUCIBILITY OF ARM VOLUME MEASUREMENTS IN THREE NORMAL CONTROLS

Measurements	*Subject*			
	S.B. Right Upper Limb	*D.D. Right*	*M.O.*	
			Right	*Left*
Number of observations	15	10	10	10
Mean hand and forearm volume ml.	1109	961	934	998.5
Standard deviation* ml.	16.4	14.1	21.4	39.1
Mean arm volume ml.	886	949.5	631.5	632.5
Standard deviation ml.	17.1	12.6	24.5	20.3
Mean upper limb volume ml.	1995	1910.5	1565.5	1631
Standard deviation ml.	24.5	14	120.5	24.2

*The standard deviations given apply to single measurements of volume.

It has been found convenient to place the tank and measuring cylinder on a wooden stand which is raised 1′ 2″ from the floor. Its overall height is then a convenient one for use with most patients. At one end of the stand a towel-rail has been added; the patient's arm is dried immediately after measurement. The apparatus is best kept near a laboratory sink.

In making measurements, a number of subsidiary but important practical points should be observed. (i) Before commencing, the cylinder base and spirit levels are checked and if necessary adjusted to the horizontal. (ii) For the patient's comfort the water in the tank should be maintained at about 70°F. The tank is filled with water and time is allowed for the overflow to cease. Greater accuracy and speed are achieved if a small

for a high degree of reproducibility in volume readings.

REPRODUCIBILITY OF READINGS

Possible sources of error in using the apparatus include: (i) Movement of the stand for cleaning purposes and failure of the observer first to adjust the level of the tank. (ii) Failure to allow sufficient time for overflow to cease, when the tank is filled before starting measurements, or when the reading is being made after immersion of the limb. (iii) Faulty position of the limb in the tank. As the patient has to bend sideways and slightly forwards with increasing depths of immersion there is a tendency for the limb to be carried away from the rule. In volume measurements of the whole upper limb, especially, this may result in falsely high readings being obtained.

The effect of such sources of error was examined by carrying out the series of measurements in three healthy control subjects given in Table 1. In subjects S.B. and D.D. only the right upper limb volume was measured, but in subject M.O., who was left-handed, the volume of both limbs was recorded.

It is apparent from Table 1 that, with the present method, hand plus forearm measurements are relatively more reproducible than are arm measurements. The standard deviation for repeated measurements of the volume of the whole upper limb in the three subjects ranged from 14 ml. to 24.5 ml. This variation is relatively small in relation to the total volume measured and to the changes to be anticipated in disease. Therefore, the degree of accuracy of the method appears to be quite satisfactory for clinical work.

renal, breast, or other disease likely to affect the volume of the upper limb.

It was desired to establish the correlation between volume and circumferential measurements of the limb at different levels, the normal relationship between hand and forearm, and arm volumes, the normal range of asymmetry between the right and left limbs, and the effect of right and left-handedness. Clearly such knowledge may be helpful in assessing the extent and distribution of limb swelling under pathological conditions.

The circumference of each upper limb was measured at three levels, at the wrist (immediately above the lower end of the ulna), at about mid-forearm level (5 inches below the tip of the olecranon) and at about mid-arm level (7 inches above the tip of the

TABLE 2

VOLUME OF UPPER LIMB AND ITS SEGMENTS IN CONTROLS

Subjects	*Number measured*	*Volumes in ml.*								
		Hand plus forearm			*Arm*			*Total upper extremity*		
		Right	*Left*	*R—L*	*Right*	*Left*	*R—L*	*Right*	*Left*	*R—L*
Right-handed controls	91									
Mean		1140	1097	43	884	870	15	2024	1967	58
Range		790 to 1685	770 to 1580	−100 to +180	400 to 1550	385 to 1575	−180 to +130	1205 to 3195	1155 to 3155	−180 to +215
Standard deviation				50			49			68
Left-handed controls	5									
Mean		1010	1031	−21	636	654	−18	1646	1685	−39
Range		900 to 1110	965 to 1070	−65 to +40	615 to 690	620 to 675	−50 to +20	1515 to 1800	1630 to 1745	−115 to +60
Standard deviation				22			28			69
Ambidextrous controls	4									
Mean		1139	1089	50	985	1026	−41	2124	2115	9
Range		920 to 1405	880 to 1245	0 to +160	660 to 1550	650 to 1695	−145 to +10	1580 to 2955	1530 to 2940	−15 to +50
Standard deviation				76			70			31
All controls	100									
Mean		1133	1094	40	876	865	11	2009	1959	51
Range		795 to 1685	770 to 1580	−100 to +180	400 to 1550	385 to 1695	−180 to +130	1205 to 3195	1155 to 3155	−180 to +215
Standard deviation				50			50			71

UPPER LIMB VOLUMES IN ADULT WOMEN

Measurements of both upper limbs have been made in 100 control subjects, all healthy adult women over the age of 30 years. Most were members of the lay staff of the hospital, of the ladies' auxiliaries, or of regional bowling clubs. As far as could be determined none suffered from significant cardiac,

olecranon)§. The volume of each upper limb, and of its two segments, hand plus forearm, and arm, was then determined as described.

§The measurements at the second and third levels mentioned are referred to hereafter as forearm and arm circumferences.

As shown in Table 2, the volume of the upper limb to maximum depth of immersion in a tank of the type described is approximately 2 litres, and in right-handed subjects the average volume of the right limb is about 50 ml. greater than that of the left. It is likely that the dominant hand, being the more used, is associated with slightly better muscular development of the corresponding limb.

tent of the swelling as follows: the volumes of both upper limbs and their segments are measured, and the values for the normal side are deducted from those for the swollen limb. The figure obtained should then be corrected by allowing for the normal range of asymmetry and the patient's handedness, e.g. applying a correction of plus 180 ml. to minus 215 ml. to the difference between the volume of the two limbs if the right limb is

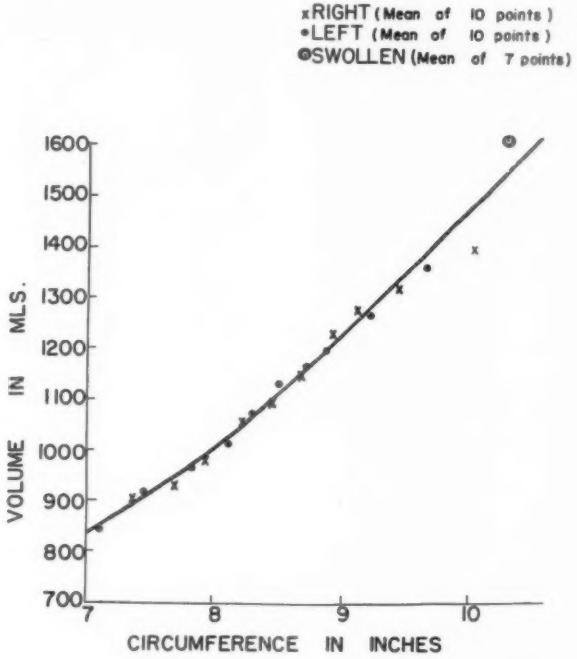


FIG. II. Circumference at mid-forearm level plotted against volume of the corresponding forearm and hand in 100 normal subjects and 7 patients with post-mastectomy lymphoedema.

In considering individual patients, the most certain determination of the presence and extent of limb swelling will be obtained by serial measurements comparing the two upper limbs before and after the onset of the pathological process. This can readily be done for post-mastectomy lymphoedema by routinely measuring all patients with breast cancer before and at regular intervals after operation. Unfortunately where the pristine limb volume is unknown as in patients first presenting with established swelling, only an estimate can be given as to the absolute extent affected in a right-handed subject. On the other hand, in comparing groups of subjects, determination of the average absolute difference between the two limb volumes, with allowance for handedness, using its average effect on volume, should suffice.

CORRELATION OF CIRCUMFERENCE AND VOLUME OF LIMB

The correlation between the circumference and volume of the two segments of the upper limb is shown in Figs. II and III, in which

are plotted the means for groups of cases in order of increasing circumference. Clearly the correlation between limb circumference and volume is very close. The swollen limbs were in patients who had previously undergone radical mastectomy for breast cancer and, as a group, they showed increases in hand and forearm, and in arm volumes lying close to the same regression line as relates volume and circumference in normal women.

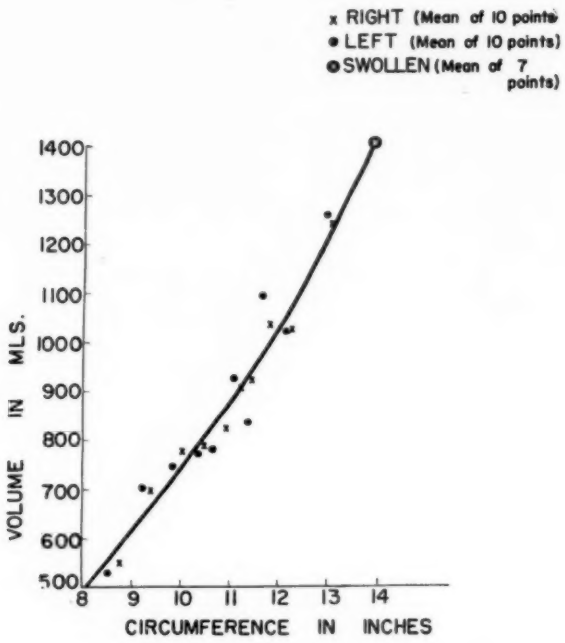


FIG. III. Circumference at mid-arm level plotted against the volume of the corresponding arm in 100 normal subjects and in 7 patients with post-mastectomy lymphoedema.

The difference in ml. between the volumes of the upper limbs has also been related to the circumferences in inches of the forearm and arm at the levels described. The regression equation expressing this relationship, as derived from our data, is as follows:

$$Ve_R - Ve_L = k\,(Cf_R + Ca_R - Cf_L - Ca_L)$$

The constant, k equals 134 ml. per inch and Ve is the volume of the upper limb, Cf, the forearm circumference, and Ca, the arm circumference. Thus from measuring the forearm and arm circumferences, the difference in volume of the two limbs can be estimated.

EXAMPLES OF CLINICAL APPLICATION

Case 1

G.W., aged 63, a right-handed married woman, stated that, before migrating from England to Australia, she had undergone an operation for removal of the left breast in May, 1951 (she had noticed a lump beneath her left nipple). As far as she knew, the operation wound had healed well; she was discharged from hospital within 3 weeks. No X-ray treatment was given to the area at any time. About 1-2 months after the operation the left upper limb was first noticed to be swollen and, with fluctuations, this swelling had gradually increased since (Fig. IV).

On 24th April, 1957, examination of the left pectoral region showed the findings usual after radical mastectomy. But there was no clinical evidence of local recurrence or of general spread of breast cancer. Radiological examination of the thorax revealed no mediastinal or pulmonary involvement.

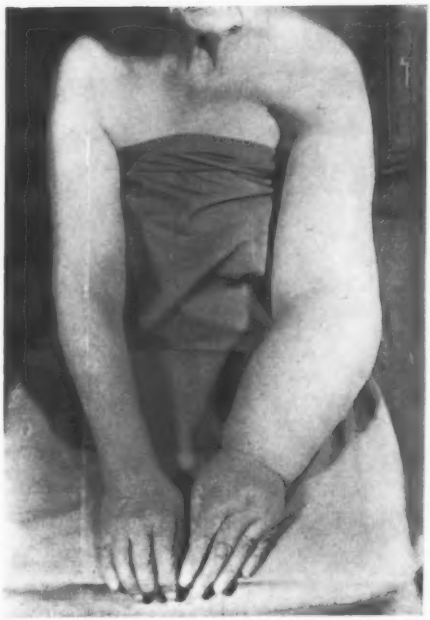


FIG. IV. Oedema of left upper limb following radical mastectomy (Case 1).

The volumes in ml. of the two upper limbs and of their segments were as follows:

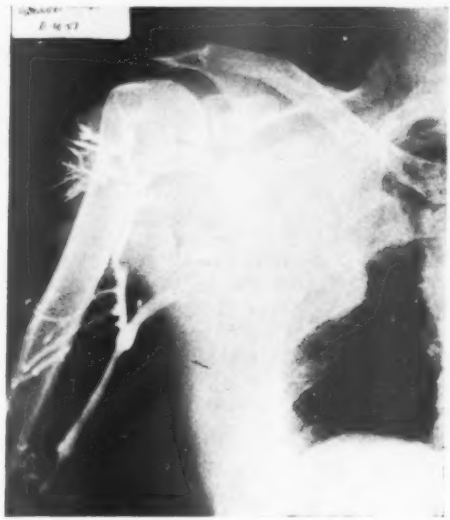
	Forearm and Hand	*Arm*	*Whole Limb*
Left upper limb	3,200	2,175	5,375
Right upper limb	1,260	1,140	2,400
Difference (L-R)	1,940	1,035	2,975

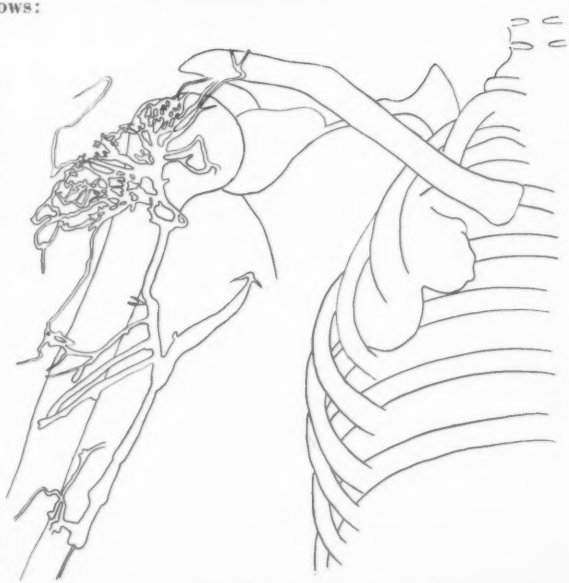
Comment. The volume of the swollen left upper limb was more than double that of its normal fellow of the opposite side and the degree of swelling of the forearm was relatively greater than that of the arm.

Case 2

R.L., a right handed male, aged 61, stated that, following a miniature mass X-ray examination in 1955, he had been told that a tumour was present in the upper part of his chest on the right side. In September, 1955, biopsy showed the tumour to be a chondroma.

Examination in April, 1957, revealed a very large and fixed tumour in the right upper chest wall with its centre below the middle of the clavicle and at the level of the second rib. There was conspicuous development of the venous collateral channels in the right pectoral and deltoid regions. Radiographs showed a large non-calcified tumour centred on the right second rib and extending back from near its anterior end at least to the mid-axillary line. The





FIGS. V and VI. Photograph of phlebogram and tracing of axillary vein obstruction by chondroma (Case 2).

first, but not the third rib, could be seen separate from the tumour.

From the prominence of the superficial collateral veins it seemed probable that the axillary vein was obstructed by the tumour mass and this was confirmed by phlebography (Fig. V.). Opaque medium, 20 cc. of 60 per cent. urografin injected into the upper part of the basilic vein through a cardiac catheter, was arrested at mid-axillary vein level. Filling of the collateral venous channels is well seen.

Measurements of the right and left upper limbs gave results as follows:

	Forearm Volume	*Arm Volume*	*Whole Limb Volume*
Right upper limb	1,990	930	2,920
Left upper limb	1,800	890	2,690
Difference (R-L)	+190	+40	+230

Comment. Despite complete blockage of the axillary vein demonstrated radiographically, there was only a slight increase in the volume of the right upper limb especially if allowance is made for his right-handedness.

SUMMARY

1. An apparatus and method are described for measuring the volume of the upper limb by water displacement. The technique enables an accurate record to be kept of volume changes in the whole limb or in its component parts, hand, forearm and arm.

2. To provide background data for studies of post-mastectomy lymphoedema, the volume of the upper limb has been measured in 100 healthy women over the age of 30 years. Its volume is approximately 2 litres and, probably because of right-handedness, the average volume of the right upper limb is about 50 ml. greater than that of the left.

3. There is a close correlation between the circumference and volume of the normal and swollen forearm, and arm. A formula is given for calculating the difference in volume of the two upper limbs from simple measurements of the forearm and arm circumference at specified levels.

4. Examples of the clinical application of the technique are described.

THORACIC AORTIC ANEURYSM AND LUNG SUPPURATION

By Rowan Nicks

Sydney

AMBROISE PARÉ in 1564 was aware of the association of syphilis with aortic aneurysm and of the gloomy progression to rupture, for he recorded the following observations. "The Aneurismes which happen in the internal parts are incurable. Such frequently happen to those who have had the unction and sweat for the cure of the French Disease, because the blood being so attenuated and heated therewith, cannot be contained in the receptacles of the artery, it distends it to that largeness as to hold a mans fist; Which I have observed in the body of a certain Taylor who had an Aneurisma of the Arterious vein suddenly whilst he was playing at Tennis fell dead, the vessel having broken; his body opened I found a great quantity of blood poured forth into the capacity of the Chest, but the body of the Artery was dilated to that largeness I formerly mentioned, and the inner coat thereof was bony."

The purpose of this brief communication, based on limited experience, is to draw attention to thoracic aneurysm as an unusual cause of respiratory symptoms which are amenable to surgical relief by different techniques best suited to the age and clinical condition of the patient and to the site of the lesion.

In five cases within my experience (one limited to the ascending aorta; one extending from the ascending aorta and arch to the descending aorta and three situated at the junction of the arch and the descending thoracic aorta) pneumonitis from bronchial compression provoked symptoms leading to diagnosis and to treatment. In the other case, included to complete the series, pulmonary symptoms were secondary to intra-mediastinal and intrapleural leakage of a mycotic aneurysm leading to pulmonary compression.

Four of the six cases were in a rapidly deteriorating and toxic physical state, with loose cough, purulent expectoration, a rapid pulse and a fever (mild in two, hectic in one) following segmental or lobar pulmonary collapse with retention of secretions which did not respond to antibiotic treatment.

Surgery, namely palliative treatment in two cases, pneumonectomy and local support in two cases, resection and homologous graft in two cases, was undertaken at this stage, the operations being limited as far as possible to the particular problems encountered.

There were two deaths, one following initially successful resection and grafting of the mycotic aneurysm from secondary haemorrhage on the tenth day and one from cerebral thrombosis, renal failure and generalized arteriosclerosis three weeks following a palliative procedure.

Case Histories

Case 1

Male (H.L.), aged 49 years. Painter.

Diagnosis. Diffuse fusiform syphilitic aortitis of the ascending aorta with local sacculation and compression of the superior vena cava and the right upper lobe bronchus.

History. On admission he complained of progressive weakness, severe cough with mild pyrexia and sweating. The distended neck veins and the obvious venous network on the anterior chest wall indicated mediastinal compression.

Fluoroscopy revealed partial collapse of the upper lobe of the right lung and a pulsatile mass in the superior mediastinum.

At thoracotomy the adherent right upper lobe of the lung and the superior vena cava were compressed.

The lesion was not considered suitable for resection and a local palliative operation was devised (Fig. 1) to protect the lung and to buttress the aneurysm against impending perforation into the superior vena cava. After freeing the lung and bronchus, the apical pleura was dissected downwards, folded around the aorta and sutured in position as a cylindrical bag.

It formed a tubular sandwich patch about the region of the ominous bulge in the dilated aorta, the filling being made with inserted blood clot and traumatized muscle. It was hoped that this would organize as a firm fibrous support. However, a gloomy prognosis was envisaged.

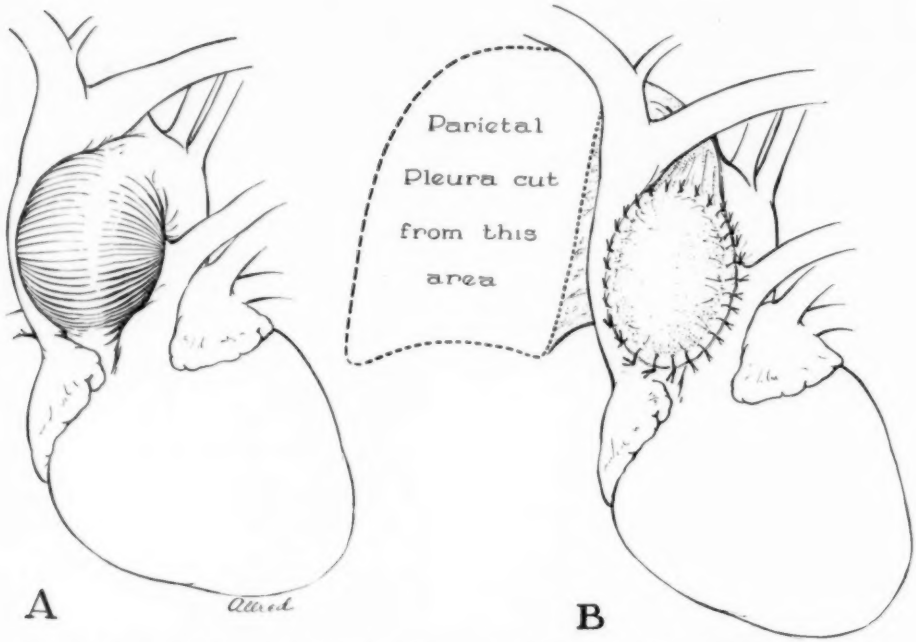


FIG. I (Case 1). Aneurysm of the ascending aorta. The ominous bulge has been supported by a pleural envelope (B).

His convalescence was uncomplicated and the pulmonary symptoms disappeared. He was given a full course of anti-syphilitic treatment and remained reasonably well, apart from angina of effort, until just before his sudden death three years later.

Unfortunately, a post-mortem examination was not obtained, but it was thought that he died from heart failure.

Comment. It is unlikely that more benefit would have followed excision and it seems that the simple palliative strengthening of the aortic wall, while the syphilitic disease itself was controlled, achieved a limited objective.

Case 2

Male (H.M.), aged 54. Retired engineer.

Diagnosis. Aortic aneurysm with compression of the trachea and left main bronchus.

History. He was admitted with cough and purulent sputum which had peristed following an accident six months previously, at which time he had coughed up large quantities of blood.

On examination there was some distension of veins of the left chest and back, and a pulsatile mass was felt in the left supra-clavicular region. The X-ray showed a large mass with calcification in its wall arising from the left hilar region and extending upwards from the left hilar region towards the thoracic inlet. An X-ray taken six years previously, had shown a small calcified aneurysm at the hilum.

At operation, performed by a sternal split incision and extending into the fourth left interspace, a great aneurysmal mass, completely adherent to the lung, was found extending up to the dome of the left pleura. The formidable tumour was freed and examined.

The original sacculation had enlarged upwards and medially behind the origin of the innominate and carotid arteries. The frail condition of the patient did not warrant radical surgery which was therefore limited to palliative dissection and wrapping with Ivalon.

He died three weeks later with a mild cerebral thrombosis.

At post-mortem examination (Dr. S. Williams) there was bilateral bronchopneumonia and tracheobronchitis, advanced generalized arteriosclerosis with complete renal cortical destruction from advanced nephrosclerosis, a small cerebral artery thrombosis involving the left basal ganglia.

The aneurysm was seen to take origin from the aortic arch and to be demarcated into two separate

masses by the vagus nerve. It was filled by laminated blood clot, and while the orifice, in the region of the left subclavian artery was only 3 cms. in diameter, the sac itself extended upwards posteriorly and enveloped the origins of the innominate, common carotid, and left subclavian arteries, all of which were atheromatous but patent. The surface of the aneurysm had been wrapped in Ivalon sponge which was completely incorporated in a fibrous reactive envelope. Aortic sections confirmed this striking infiltration of active vascular fibroblastic proliferation into the interstices and maintained the continuity of the aorta (Fig. II).

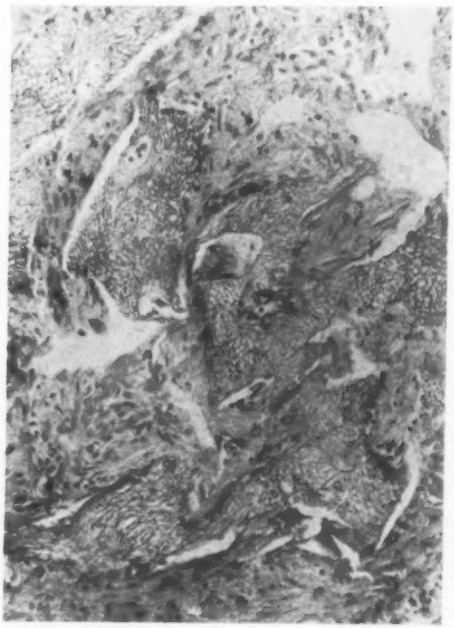


FIG. II (Case 2). Showing Ivalon sponge infiltrated with fibrous tissue granulation three weeks after wrapping of aneurysm. There is no inflammatory foreign body reaction.

Comment. This case is illustrative of the limitations of major surgery by age and general disease, the difficulty of prognosis in individual cases, and of the efficacy of Ivalon wrapping as a minor palliative procedure.

The aneurysm, diagnosed six years previously, had caused no inconvenience during this period. If resection had been possible a this time, survival might have been attributed to successful excision. Such delay in treating young persons with a traumatic aneurysm, by allowing progression almost to inoperability, would be inexcusable.

Case 3

Male (C.P.), aged 56. Retired civil servant.

Diagnosis. Aneurysm of the aortic arch and first part of the descending aorta. Suppurative pneumonitis; pneumonectomy and pericardial wrapping.

History. He was admitted complaining of progressive asthenia, sweating and constant loose cough with purulent expectoration. Temperature 101.5° F; pulse 130/min.; W.B.C. 16,800 of which 73 per cent. were segmented forms. The chest X-ray showed diffuse pneumonic changes at the left base and a large irregularly calcified mediastinal mass (which was later recognized by fluoroscopy to be an aneurysm of the ascending aorta, the aortic arch and the descending aorta) compressing the left main bronchus. His condition deteriorated rapidly and soon became critical despite the administration of large doses of suitable antibiotics. After pondering the problem, a left pneumonectomy was advised and performed without undue difficulty in order to remove the suppurative process distal to bronchial obstruction.

The left main bronchus was compressed to a slit which did not leak air on section but the mucosa was intact.

A large rectangular flap of the pericardium was fashioned and wrapped around the aneurysm as a protective cylinder which, however, extended beyond this across the arch.

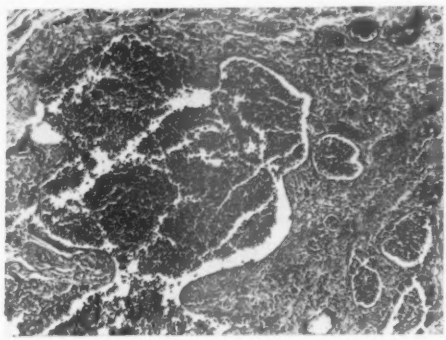


FIG. III (Case 3). Chronic suppurative pneumonitis showing the bronchi partially denuded of epithelium and distended with pus. The surrounding lung is infiltrated with fibrous tissue.

Convalescence was uneventful and he was relieved of major chest symptoms. The bronchial system of the gross specmen was distended with pus, the histological examination confirmed the presence of purulent bronchitis with collapse and infiltration of the peribronchial lung tissue by chronic inflammatory cells. The changes were non-specific and consistent with suppurative pneumonitis (Dr. S. Williams, 18th November, 1955) (Fig. III).

He died a year later from sudden rupture of the aneurysm into the trachea.

Comment. Prior to operation he was in a critical state.

Immediate improvement followed relief of the suppurative pneumonitis by pneumonectomy.

It is doubtful if the pericardial flap was a protection against the rupture of this diffuse aneurysm.

Case 4

Female (Mrs. P.), aged 58.

Diagnosis. Suppurative pneumonitis of the left lung secondary to thoracic aneurysm of the aortic arch. Pneumonectomy and palliative support of the aneurysm with Ivalon sponge.

History. She was admitted with a history of cough and purulent expectoration of six months duration. Her general condition had recently deteriorated. On examination she was toxic, thin

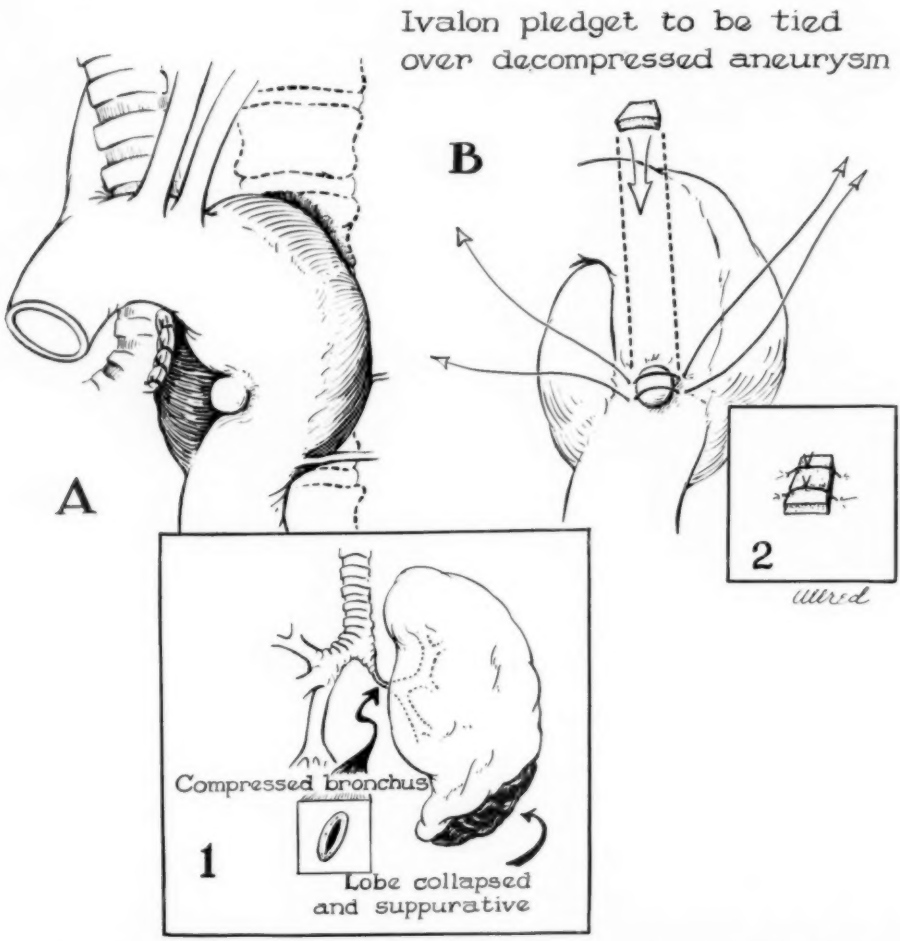


FIG. IV (Case 4). Aneurysm of the terminal arch after pneumonectomy had been performed the insert (1) showing compression of the left main bronchus with collapse of the left lower lobe and obstructive emphysema of the left upper lobe. A localized ominous bulge has been inverted in insert (2) encircled in Ivalon sponge (B).

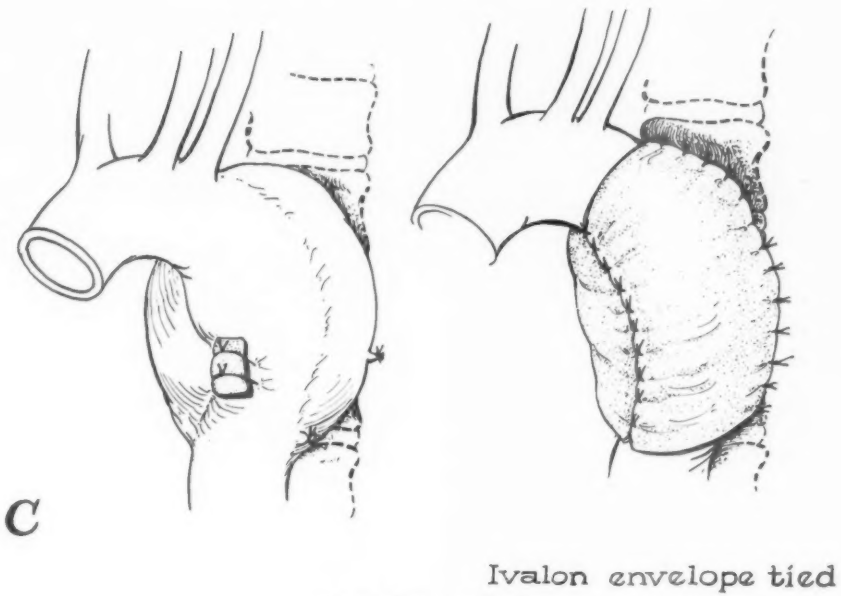


FIG IV (Case 4 continued).

and in poor condition, with a hectic temperature. She was coughing up almost pure pus. The chest X-ray showed inflammatory infiltration and collapse of the left lower lobe of the lung and obstructive emphysema of the upper lobe, associated with a mass in the left hilum (Fig. IV). This was shown on fluoroscopy to be an aortic aneurysm.

On the basis of the previous experience, pneumonectomy was performed and after the aneurysm had been freed entirely from the upper dorsal vertebrae, resection of the aneurysm, which was maximum at the level of the subclavian artery, was debated. It was decided that her general condition did not warrant this procedure. Accordingly, two small very dangerous bulges were inverted with purse string sutures, and the whole covered with a cylinder of Ivalon sponge (Fig IV).

Her convalescence was uneventful and she remains well and grafting of this aneurysm could be done safely now (two years later).

Comment. As in the previous case, pneumonectomy was life saving and while local treatment of the aneurysm has forestalled the possibility of immediate rupture, the field is clear for resection and grafting as a second stage procedure.

Case 5

Male (J.W.), aged 49.

Diagnosis. Aneurysm at the junction of the transverse and descending aorta.

History. He was admitted in good condition with a mild cough but without evidence of gross bronchial obstruction. The chest X-ray revealed a localized aneurysm in the left hilar region. At subsequent operation under hypothermia, a large saccular aneurysm extending upwards as high as the origin of the left subclavian artery was excised, and the aorta reconstituted by an homologous freeze-dried aortic graft. He has remained well, and the chest is clear.

Comment. This is the ideal type of case to withstand resection and grafting. Some form of local by-pass from the aorta above and conducting blood into the artery below the aneurysm to permit clamping of the isolated aneurysm without jeopardizing the heart or distal circulation is now preferred to hypothermia.

Case 6

Male, aged 24.

Diagnosis. Rupture of mycotic aneurysm of the descending aorta. Haemothorax and compression of the lung. Resection and grafting.

History. This Polynesian was admitted to the care of Dr. E. G. Sayers with a history of rapid decline, fever, sweats, mild jaundice and great weakness. The chest X-ray was clear at this time. He

complained of the onset of dysphagia which progressed until it became absolute. A barium swallow confirmed the presence of complete oesophageal obstruction by a mass compressing the oesophagus from without. A chest X-ray taken at this time showed a mass in the lower left chest extending from the hilar region but the diagnosis was obscure.

FIG. V (Case 6). Dying of secondary haemorrhage from an aortic graft on the ninth post-operative day following resection of a ruptured mucotic aneurysm. Secondary perforation of the oesophagus from aneurysmal pressure necrosis occurred and has broken down after treatment (shown on the left).

He suddenly developed severe pain in the chest, and at thoracotomy a thoracic aneurysm was found leaking into the mediastinum and causing massive haemothorax without compression of the lung. He was submitted to a further thoracotomy under hypothermia (cooling rapidly from 104° to 90° without harm).

The lung was found to be compressed by blood and clot in the pleural cavity coming from an aortic aneurysm one inch below the arch and leaking into the mediastinum and pleura. The aorta was freed above and below the defect restored by a homologous aortic graft. Immediate recovery was satisfactory, but on the third day spontaneous perforation of the oesophagus occurred which was explored and found to originate from localized gangrene, the oesophagus having been incorporated in the wall of the aneurysm. The gangrenous area was excised, and the oesophagus locally reconstituted. He perished from secondary haemorrhage following rupture of the graft on the ninth post-operative day (Fig. V). Section of the aneurysmal wall confirmed the mycotic nature of the aortic lesion, and a pure culture of haemolytic staphylococcus aureus was grown.

Comment. He was unable to give any account of himself.

There was a vague possibility that he might have swallowed a fish bone but none was seen at operation or at a careful post-mortem examination. The operation presented no great technical difficulties. It was unfortunate that the oesophagus had been devitalized by the aneurysm. This close relationship is important. His name was Tusitala, Teller of Tales. He awakened from coma during the induction of hypothermia, saw a mountain of ice over his body, opened his eyes very widely and then shut them tightly while he was given his intravenous Pethidene and oblivion.

DISCUSSION

Very little attention, medical or surgical, has been focussed on the occurrence of obstructive pulmonary suppurative complications, resistant to medical therapy, as a cause of decline and death in thoracic aneurysm.

This is surprising in view of such post-mortem studies as those of Boyd (1924), Kampmeier (1938), and Brindley and Stembridge (1956), all of which showed this to be the dominant pathological lesion in ten per cent. of the cases.

Of 201 cases of thoracic aortic aneurysm admitted to the Royal Prince Alfred Hospital from 1910-1949 (165 males, 36 females at a mean age of 53 years), the duration of symptoms of those dying in hospital was 16 months.

Syphilis, contracted at an average of 25.6 years, previously was the proven (WR positive) predisposing cause in 63 per cent. of cases.

A brief review of the 46 cases dying in hospital revealed that the aneurysm involved

the aortic arch in two-thirds of the cases, one-third of these being combinations of the arch with the ascending and descending aorta. The causes of death in these cases (tabulated below) are in general accord with the larger statistics already mentioned.

ROYAL PRINCE ALFRED HOSPITAL

DEATHS FROM THORACIC AORTIC ANEURYSM, 1910-1949

Complication	*Site*	*Total*
Rupture	(a) Bronchus .... 5 (b) Pleura .... 5 (c) Pericardium .. 2 (d) Oesophagus .. 2 (e) Elsewhere 2	10
Congestive Cardiac Failure	——	12
Pressure on Trachea or Bronchi	Infection not commented upon .... 5 Broncho-pneumonia 5	10
Other causes	——	8
	TOTAL:	46

Many cases of aortic aneurysm have become amenable to surgical treatment (Bahnson, 1953; Cooley and De Bakey, 1955). The position and extent of the lesion may be visualized by aortography and satisfactorily performed by injecting 70 per cent. Diodrast directly into the left ventricle (Lehman, Musser and Lykens, 1957). In most cases operability is assessed at thoracotomy and is planned in accordance with the clinical condition, especially the cardio-renal function. A saccular aneurysm suitable for this type of treatment is excised and the aorta sutured through firm tissue beyond the occluding clamp (Bahnson, 1953). Larger and more diffuse aneurysms of the distal arch and descending aorta, provided the patient is in a fit state to withstand the procedure and that an occluding clamp can be applied beyond the left common carotid artery (Cases 5 and 6) are best cured by excision and grafting, care being taken to protect the delicate nervous cells of the spinal cord during this period by one of the following techniques:

(1) by hypothermia (De Bakey, 1954; Nicks, 1955);

(2) by an arterial shunt from the left atrium to the femoral artery (Cooley, De Bakey and Morris, 1957);

(3) by using the pump oxygenator, the venous blood being withdrawn from the vena cava oxygenated and returned to the femoral artery.

The last two techniques have been developed to prolong the safe time limit for aortic occlusion and to relieve cardiac strain in cases in which the danger of cardiac collapse is envisaged.

While homologous grafts are in general use, plastic prostheses (Edwards-Tapp, *etcetera*) are being used increasingly and with confidence. Recently the operative field has been extended to include the aortic arch itself. However, the mortality for heroic surgery in thoracic aneurysm is in the neighbourhood of 30 per cent. in the best hands (De Bakey and Cooley, 1957). Palliative treatment, although not ideal, does permit intolerable symptoms to be relieved and life to be preserved. A left pneumonectomy by itself, with or without support of the weakened wall of the aneurysm by Ivalon sponge or living tissue is immediately life saving in the very sick, especially when the lesion is unsuitable for local resection and grafting. If the local lesion is suitable for grafting, this can be done at the same time, or at a later date depending largely on individual circumstances. Occasionally local palliation may have a place in the treatment of ascending and aortic arch aneurysm unsuitable for local resection where this is not in the best interests of the patient. In selected cases, local support of the vessel with Ivalon sponge is recommended, occasionally combined with local excision or inversion, as providing a fibro-elastic support which does not interfere with the nutrition of the tunica media. The use of reactive cellophane has been discarded, for it has been found to exclude the vessels from the tunica adventitia and so interfere with the nutrition of the tunica media, that further weakening of the

wall occurs. Of twelve in a series of cases of abdominal aneurysm so treated at the Green Lane Hospital, Auckland, by this method (combined with various palliative procedures such as partial ligation of the aorta just proximal to the lesion) performed before resection and grafting was practised, there was eventual progression to rupture.

The plastic technique described (Cases 1 and 2) using pericardium or double pleural envelope containing blood clot, may be helpful in circumstances where the need for palliation is unexpectedly encountered.

ACKNOWLEDGEMENTS

Much of the work presented was done with my friends and erstwhile colleagues at the Green Lane Hospital, Mr. Douglas Robb and Dr. S. Williams. The scrutiny of the Royal Prince Alfred Hospital records has been possible only with the help of my registrar, Dr. Hinde.

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METASTASIZING BASAL CELL CARCINOMA

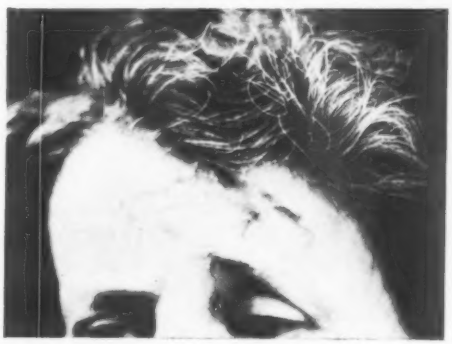
A CASE REPORT

By B. K. RANK

Melbourne

THE two main variants of epidermoid carcinoma, squamous cell and basal cell lesions, apart from their characteristic histological features, demonstrate sufficient difference in their clinical behaviour to warrant this commonly accepted basis of classification. These differences were in fact well known and described (Jacob, 1827*) long before the use of the anacromatic microscope and the introduction of staining procedures.

By far the most striking of these differences in behaviour concerns their liability to metastasize. The rarity of metastasis from basal cell carcinoma poses something peculiar in view of a cellular picture in which mitotic pictures are so common. Various hypotheses are raised to explain this oddity — including observations that it is inter-cellular adhesion or the integrity of the palisade layer which prevents dissemination of free cells from the growing mass of a basal cell carcinoma.



A

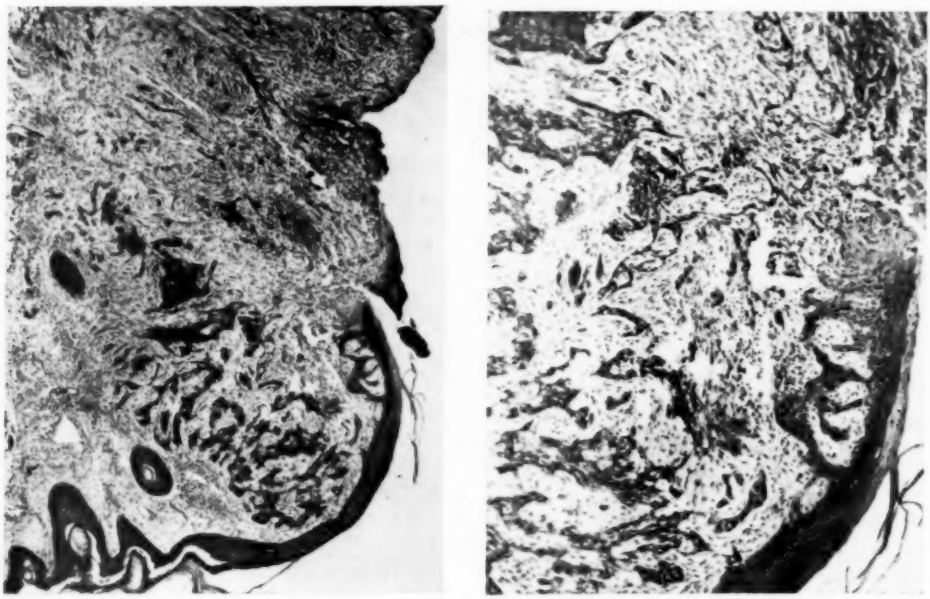
B

FIG. I

*Arthur Jacob (1790-1874) was a very distinguished Irish surgeon. President of the Irish College of Surgeons in 1837 and again in 1864-65. A student of Colles, he became a distinguished teacher himself but was best known as an anatomist and an ophthalmic surgeon. He wrote a classic textbook of ophthalmology and devised many instruments. Among his many anatomical discoveries was that of the retinal layers of rods and cones. His was the first recorded description of what is now called "rodent ulcer."

It is well known, however, that the absence of metastasis from a basal cell carcinoma is by no means a hard and fast rule. Mixed lesions (basi-squamous) demonstrating histological features of both squamous and basal cell carcinoma are by no means uncommon. A few prickle cells and cell nests are often noted in basal cell lesions if one is actively looking for them and so pathologists differ in what they would term a mixed lesion. In

a recent review of over 500 cases of cutaneous carcinoma by the writer and his associates 23 truly mixed and indeterminate lesions were noted (5.5 per cent.). From these mixed lesions metastases may occur. Furthermore, we have seen typical examples of basal cell carcinoma in course of time change their morphology and behaviour, becoming typical report. Case reports of this condition are apparently arise from squamous elements. Metastasis of typical basal cell carcinoma as basal cell carcinoma does however occur but is sufficiently rare to warrant the ensuing case Finnerud in 1924 described two cases with lymph node infiltration. Singer in 1934 had one fatal case with widespread skeletal metastasis as well as lymph nodes. De Nevasquez in 1941 made record of another fatal case



A B

FIG. II

very few. Some of these are insufficiently documented to merit consideration, while others are clear examples of squamous cell metastasis from mixed lesions (as one reported by Hunt, 1952) or lesions which have changed their nature. One has been able to sight only five acceptably reported examples of pure basal cell metastasis. All were from primary lesions on the face. Beadles in 1894 described without illustration a single sub-maxillary lymph node with basal cell infiltration, discovered at post-mortem from an extensive primary lesion on the face. squamous carcinoma. In either of these circumstances if metastases occur, they are squamous cell or mixed in nature and with widespread involvement of bones and the lungs. The case reported here is perhaps unique in the outcome of treatment.

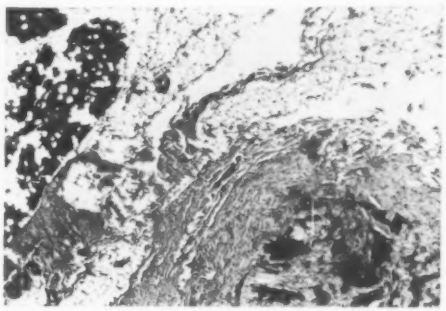
Case report

Mrs. S.C.I., age 49, was first seen by the writer in December, 1946. She gave a history as follows.

Twelve years previously (1934) when aged 37, following a vague history of injury, there was "a thickening" on her forehead which was regarded as an excess of scar. This was excised, following which she was treated with superficial radium and healing occurred.

Eight years previously the area had broken down to form an ulcer on the left side of her forehead which in subsequent years slowly got bigger despite varied treatments including X-ray therapy. The ulcer became painful and during the last year extension was more rapid and bleeding was frequent.

When first seen in 1946 the condition was that illustrated in Fig. I (a). There was an ulcer on the left side of forehead extending from hair line to eyebrow which had typical clinical features of a large rodent ulcer. There were no palpable glands in the pre-auricular or neck regions. At operation (I) (December, 1945), a radical excision of the region was undertaken. This included removal of the outer table of the skull beneath the ulcer and the whole thickness of skull beneath its central area. Primary repair of the large defect was carried out using a direct flap from the forearm. The donor site was dressed with a split skin graft and posture of arm to forehead was maintained by plaster.



A

FIG. III. Section taken from mass beneath the ear.

(a) Shows basal cell metastasis in fibrous tissue in region of parotid gland.

(b) Further section from same specimen with basal cells in nearby lymph gland.

(c) High power of this lymph gland.

At operation (II) three weeks later, the flap based upward on the forehead was detached from the forearm. There have been no further operations to the forehead region.

The pathologist (Dr. A. V. Jackson) reported on the tissue excised as follows:—

"Bone and soft tissue from forehead: Edges and most of the base of the ulcer showed typical basal cell carcinoma" (Fig. II [a] and [b]).

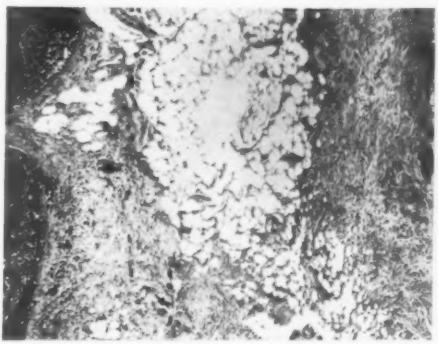
A report on the bone followed:—

"Bone shows some chronic inflammatory changes only, there is no histological evidence of neoplastic invasion, the bone proved to be harder than expected and had to be passed through the decalcifying fluid a second time."

Convalescence was complicated by femoral thrombosis and a swollen leg which gradually improved and the patient left hospital for her home in another State (15th February, 1947).

During a routine review six months later a small hard mass was first noted beneath the lobule of the ear. As this continued to enlarge she was re-admitted to hospital and the mass removed for biopsy on 25th November, 1947. This time the pathologist reported:—

"Neck: Gland is invaded by carcinoma cells which are darkly staining resembling the basal cells of the skin rather than the more superficial squamous cells. There is no doubt now, of course, that the tumour is malignant and invasive. I still consider that the original lesion was a 'basal cell carcinoma,' but it has now developed the invasiveness usually associated with a squamous cell carcinoma" (Fig. III).



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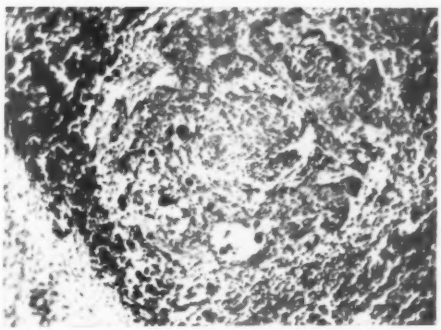

C

One week later, 2nd December, 1947, a limited dissection of the upper neck gland field was carried out. It was certainly not a radical and complete neck dissection. The pathologist reported:—

"Glands of the neck for section: Several small glands are invaded by a metastasizing basal cell carcinoma. Other lymph glands in the block of tissue appear to be free from carcinoma."

A radiotherapist advised against radiotherapy at this stage. Six months later, however, there was a palpable gland in the left posterior triangle of the neck and after consultation interstitial radium to the

neck was advised and carried out by Dr. Kaye Scott. Following this rescission of the mass occurred. She has been seen at regular intervals since that time and though there is some thickening and fibrosis in the neck, there has been no change over the years and there are no palpable glands. Her condition nine years after her last treatment is shown in Fig. I (b).

Comments

(1) As will be seen from the histological picture of the ulcer and the glandular metastases, this case is clearly one of the rare examples of a metastasizing basal cell carcinoma.

(2) It demonstrates, too, the pitfalls of a rigid outlook, regarding prognosis and management, based on a histological diagnosis without proper regard for vigilance on the gland field.

(3) It demonstrates a common inadequacy of a gland field dissection for upper face regions which does not include a full dissection of the posterior triangle of the neck.

(4) It demonstrates both good and bad in the use of both surgery and radiotherapy. Though the latter is not generally regarded as a satisfactory therapeutic agent for malignant glands of the neck, it has in this case been an outstanding success.

(5) The patient is very much alive and well ten years after her first operation and the order of reconstruction achieved for a hideous defect of the forehead speaks for itself (Fig. I [b]).

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HYPOTENSIVE ANAESTHESIA FOR PLASTIC SURGERY

By John Tucker

Melbourne

THE specialized field of plastic surgery demands in common with neurosurgery a high degree of technical precision and it is no wonder therefore that the control of bleeding is of considerable importance as a factor contributing to the actual standard of surgical result. While that part of plastic surgery performed on the limbs is usually done under a tourniquet for this very reason of surgical precision, the bloody field of facial surgery has always presented a challenge.

It is unfortunate that the use of ganglion blocking agents to reduce this bleeding has fallen into disfavour in so many departments due mainly to reports of fatal results. Good results from hypotension are comparatively easily obtained in the head and neck region and the advantages are so great, that more use should be made of this valuable part of anaesthesia.

Dr. L. G. Travers has been kind enough to allow me to include his cases in this series to make a total of 125 plastic operations on the head and neck region.

Advantages

The surgical conditions fall into several categories. The patients in whom the indications are strongest are those requiring extensive excisions for malignancy. The advantages here are considerable and can make a great difference to the ability of the surgeon to achieve a good result. For example, the outward appearance of a basal cell carcinoma does not reveal the extent it has burrowed, so that the surgeon finds it necessary to plan wide excision in all dimensions and through any structures in order to assure complete excision. When this is necessary among the crowded areas of eyes, nose, mouth and ears, a small part of an inch can make the difference between intolerable deformity and a reasonable cosmetic result. With the tissues rendered almost bloodless he can execute with much greater precision an adequate excision. Also the recognition and preservation of important structures during dissection is made easier, for example, avoiding branches of the facial nerve while removing a parotid gland or the preservation of a tear duct or recurrent laryngeal nerve.

It is fortunate that in this group where the indications are strong, the members are usually elderly patients and the anaesthetic risk possibly greater. A second group is found among younger patients and here the advantages are not so great. These are excisions of large surface areas such as excision of large naevi, the remaining defect then being covered by rotation flaps or large free grafts. Reduced bleeding allows quicker and more accurate excision and preparation of flaps with less blood loss and trauma. Also the flaps are less likely to become oedematous and congested. We feel that hypotension has helped a great deal for this but there is a further stage, that is establishment of final haemostasis before the graft or flap is placed in position and dressings applied. Naturally, hypotension is a great help here too, but as will be mentioned later, reactionary haemorrhage appears to be more likely and it may be necessary to abandon its use in these cases or allow the pressure to rise after the dissection is complete.

The primary or secondary repair of cleft palate in adults is quite a different proposition from the operation in children for it is always accompanied by profuse bleeding and haemostasis before returning the patient to the ward is often difficult and very tedious. Occasionally a blood transfusion has been necessary in adults, whereas it is never necessary in infants. Hypotension is very effective in these instances, a fact a little surprising considering the posture necessary, i.e. the same as for tonsillectomy.

Operations on the nose such as excision for carcinoma, bone grafts, reduction of old fractures or forehead rhinoplasty are all

accompanied by a great deal of bleeding. On the other hand, perhaps due to the ease of posturing, hypotension in this area produces a very good result and permits a much more precise operation. Many operations of a less essential nature and purely cosmetic procedures are suitable also for the use of these drugs but we only choose the healthy and young patient for hypotension.

every one inch above the heart. Thus if the brain is above heart level then the pressure must be kept the appropriate amount higher.

On the other hand, it is most people's experience, as it is ours, that 60 mm. of mercury, brachial blood pressure gives a satisfactory operative field and it is not necessary to go below this.

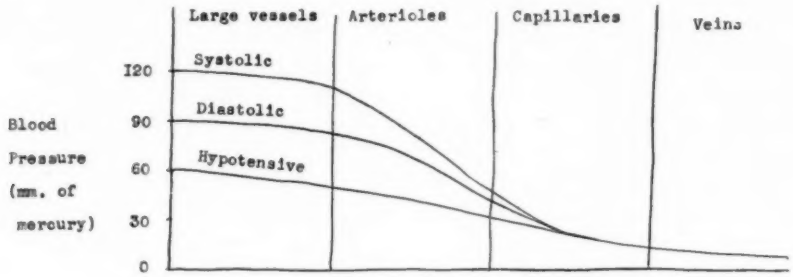

FIG. I. Distribution of peripheral resistance.

DISADVANTAGES AND CONTRA-INDICATIONS

The constant question for the anaesthetist is "should the patient be put to any added risk" and, assuming the advantages are many, it is important to assess what is the extent of the "added risk."

The risks fall into two groups, those of a serious nature due to circulatory disturbance of brain, heart, kidney, retina or liver, and those occurring post-operatively such as reactionary haemorrhage and prolonged hypotension.

Concerning the first group, it was stated by Landis (1930) that for a satisfactory circulation and tissue metabolism, the pressure in the arterioles need only be just above the sum of the pressure in the venous side of the capillaries and the osmotic pressure of the plasma, i.e. about 32 mm. of mercury.

As most of the resistance is in the arterioles, if this is removed by sympathetic block, the systemic blood pressure itself need not be much above capillary blood pressure. This statement was qualified by Griffin and Gillies (1948) who said that it is true only when the blood volume and oxygen saturation are normal and no vasoconstriction is present. Enderby and Elmore (1951) added to this again by pointing out that the capillary pressure falls .2 mm. of mercury for

Therefore using these principles if the blood volume and oxygenation are always normal, if there is no vasoconstriction, if the blood pressure is kept above 60 mm. of mercury and this figure is adjusted according to posture, then the risk of these serious visceral complications is no greater than with any other anaesthetic.

Reported figures do not help much in the assessment of risk of serious complications as there are so many variables and our own series is not nearly large enough. Enderby (1950) reported 250 cases without a death. McIndoe (1956) reported 4,500 cases of head and neck surgery in which there were 5 deaths and during the same period in 5,000 patients given no hypotensive drugs, there were only 2 deaths. The details of the deaths are instructive. In 3 cases no mechanical aid for recording the blood pressure was used and the exact level was not known for some time; this must lead to trouble. The pulse is not usually palpable at 60 mm. of mercury, nor can it be heard with a stethoscope. They state themselves that since they have used an oscillometre there has been no further trouble in this regard. The fourth death was due to air embolism, a complication for which one must always be on the look out. Recently, at the Royal Melbourne Hospital during a dissection of glands of

the neck, the assistant drew attention to air bubbles moving along the internal jugular vein with each respiration. The fifth of McIndoe's deaths occurred in a coloured man whose airway became obstructed back in the ward and was not discovered in time. It cannot be denied that the margin of error is reduced and mishaps as the above can result in death whereas without hypotension they may not occur. Thus if a patient presents any particular technical difficulties we regard this as a contra-indication to the use of hypotension.

Hampton and Little (1953) collected reports of 21,000 cases from anaesthetists and showed a death rate of 1 in 500 (twice McIndoe's rate). The deaths were due to (i) anuria, (ii) coronary thrombosis, (iii) cerebral thrombosis, (iv) cardiovascular collapse, and the rest the cause was not named. However, many of these were very sick patients. There was a high percentage of neurosurgical cases included and there was no suggestion of any choice of cases. I think that most anaesthetists agree now that any evidence of coronary insufficiency, cerebrovascular or renal disease or poor liver function is sufficient to make the use of these drugs unjustified however strong the surgical indication.

Finally, age itself is not a contra-indication. Advanced coronary disease can exist in a man of 30 while a man of 70 may have no atheroma. In fact the well preserved elderly patient is a much better risk at all times than the over indulgent patient of 40. Also it must be remembered that good results from hypotension are much more easily obtained in these patients.

Less serious complications include reactionary haemorrhage — and prolonged hypotension. Secondary haemorrhage and post-operative infection are also mentioned in some reports as being increased but we have not found any evidence of this.

Reactionary haemorrhage

Reactionary haemorrhage seems to be made more likely in certain types of operation. For example, in the 15 cases of large surface excisions and replacement of skin, there were 3 cases which developed large post-operative haematoma. It can be seen that death of a flap resulting from the haematoma would have far-reaching results. Two of these occurred in young adults who in both instances developed tachycardia and did not respond well to the hexamethonium and bleeding was not reduced very greatly. The third was an elderly patient with hypertension who responded well to the drug but bled viciously several hours later. One would expect gland dissection of the neck where large flaps are raised would also come into this category but so far in seven cases there has been no tendency to haematoma formation. This may be due to the fact that dissection lasts longer and more vessels are tied.

There was one case of haematoma among the 36 cases of excision of rodent ulcers of the face. He was a man of 65 whose blood pressure continued to fall even while the table was level so that 5 mgm. of methedrine was given intravenously before the dressings were applied. A haematoma appeared under the skin graft almost immediately. This was removed but recurred later in the ward. As Van Bergan and others have pointed out these patients are very sensitive to methedrine or neosynephrine and it should only be used in small doses and in extreme conditions.

In some other operations such as nasal reduction, bone grafts and repair of adult palates the tendency for reactionary haemorrhage was reduced.

Prolonged hypotension

There was one case that we felt came into this category. She had her pre-operative medication of omnopon gr. 1/3 and scopolamine gr. 1/150 repeated during the afternoon as the starting time for operation was delayed. As her blood pressure was still 70 mm. of mercury four hours after operation nor-adrenaline was given without very much improvement. Later 5 mgm. of methedrine was given half-hourly for two hours when the blood pressure remained above 100 mm. of mercury, unaided.

In our experience the average case returns to the ward with a blood pressure of 90 mm. of mercury and this is above 110 mm. of mercury in four hours. The patient is not

allowed to sit up until next day. Nasal oxygen or blood transfusion is given if required. Very occasionally the foot of the bed is raised but the use of pressor drugs has been avoided except for the above case.

TECHNIQUE

Pentothal, relaxant and nitrous oxide and oxygen has been used in all cases. Pentothal is used in larger quantities and often repeated early in the anaesthetic to help maintain the hypotension.

The relaxant in these cases is used solely for intubation and to assure a quiet patient while the blood pressure is being stabilized. Only a small dose is used and it is not repeated. Once the patient is under the influence of vegolysin there is no tendency for any movement and respiration is easily controlled.

Tubarine is the most satisfactory relaxant. Flaxedil makes hypotension more difficult and increases bleeding. Scoline does not last long enough for the purpose.

The hexamethonium bromide is given with the patient lying flat and the initial dose must be large — 50 to 100 mgm. If the blood pressure is not below 100 mm. of mercury a further dose is given. Then depending on the effect the patient is postured with a varying degree of head up tilt. It is the posturing that accounts for most of the fall and extreme care is taken at this stage. Although we avoid pressures below 60 mm. of mercury I make an exception here. The pressure often drops to about 40 or 30 but rises again almost immediately. Almost constant blood pressure recordings are taken at this stage and by means of increasing the tilt, or further doses of hexamethonium or pentothal the pressure is kept at 60 mm. of mercury. If this tendency for the blood pressure to rise is combated early in the anaesthetic it soon settles to a fixed level. It is very noticeable that older people have a sharper fall in blood pressure and require less drug to maintain the level of 60 mm. of mercury. In fact it is usually found that the initial dose is sufficient and posturing is only slight, a few degrees serving to maintain a constant level.

Posture

In these operations on the head and neck I feel it is important to keep the brain as near to heart level as possible. If this is not so then the blood pressure reading is kept at a higher level as mentioned above. Posture A is thus safer than B.

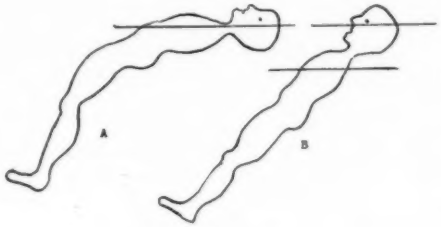

FIG. II. Posture.

Respiration

The type of respiration is very important. Controlled respiration gives the best results if carried out properly. If spontaneous breathing is allowed under ventilation will occur.

It has been shown experimentally by many people (Watrous *et alii*, 1950) that controlled respiration increases venous pressure and circulation time and reduces cardiac output. However, it is obvious that the reduced venous return only occurs during the positive phase and that the summation of interference is negligible if the bag is handled correctly.

Motley *et alii* using right heart catheterization have shown that there is no influence on the circulation if the inspiratory phase is short and the expiratory phase is long enough to allow several heart beats to occur to compensate for venous hold up.

Many anaesthetists have noted that if the pressure in the chest suddenly rises (for example if the bag becomes too full) the pressure falls immediately and becomes unrecordable. Enderby in fact has recommended using constant positive pressure to reduce the blood pressure in resistant cases. This cannot be wise as although it reduces the pressure in the large arteries it does not reduce capillary pressure or the bleeding. In practice the dryest field is obtained if some negative pressure can be applied in the expiratory phase. When possible I use a

Harrington James respirator with strong negative pressure and note that the capillary ooze is less.

Measurement of blood pressure

Many of the fatal cases described in the literature give a picture of the anaesthetist being unable to record a blood pressure and then feeling femoral and carotid artery regions in an attempt to feel a pulse. When at last it is discovered there is not a pulse it is too late. Enderby and MacIndoe both state that since they have had mechanical means to determine a pulse, no troubles at all have arisen. If a sphygmomanometer cuff is carefully placed over the brachial artery and bandaged on firmly with a non-elastic bandage, then pressures as low as 20 mm. of mercury can be shown by the flick on an aneroid manometer. The drugs should not be used without some such aid.

Duration

Most of our operations last about an hour and a half. However, some are up to three hours. Two gland dissections lasted five hours. On these occasions and also in forehead rhinoplasty and other lengthy procedures, after about two hours and when convenient the patient was put back to level position. Oozing would occur and the larger vessels would be clipped and tied. After about 15 minutes the original posture would be resumed and the blood pressure fall to about the same level as originally. This was repeated again in about an hour on several occasions.

Blood transfusion

Most plastic operations only deal with surface structures and there is no chance of bleeding from very large vessels. Thus most of the operations were done without an intravenous drip set up. However, for any of the longer procedures blood was cross typed and a drip set going.

CONCLUSION

It is our experience that so long as the definite limitations and criteria mentioned above are adhered to, the patient is not exposed to risk of any serious complications by the use of hypotensive anaesthesia. Where catastrophies have occurred in the past, there has been some wide variant from these rules and it seems important that these instances should not result in the abandoning of this type of anaesthesia.

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URETEROCELE IN INFANTS AND CHILDREN

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URETEROCELE in children is not common, but it cannot be said to be rare. It may be discovered in the course of routine urinary tract investigations for pyuria, or it may appear dramatically as a red fleshy mass at the urethral orifice.

Ericsson (1954) has reviewed very thoroughly the literature on this subject and has described in detail the clinical, radiographic and pathological features and treatment of ectopic ureterocele in infants and children, from his own experiences. He also described the congenital and acquired anomalies associated with this condition in other parts of the urinary tract.

However ureteroceles in children have special dynamic features, some of which have a definite bearing on one's plan of surgical treatment. The significance of these features needs further emphasis.

In this paper the results of a study to determine the surgical implications of these characteristics are described.

This knowledge enables the surgeon to anticipate complications which emanate from a ureterocele after the corresponding kidney has been excised. Definitive surgery can be so designed to forestall most of these complications arising from both the defunct kidney and the ureterocele.

Ureterocele is a term applied to a globular dilatation of the ureter in its course in the submucous layer of the base of the bladder.

In preference to the more complete and complex classifications of Campbell (1951), Gross and Clatworthy (1950), and the anatomical classification of Ericsson (1954), ureteroceles are divided, in this paper, according to the type of obstruction. There are two groups (Fig. 1):

(1) The stenotic ureterocele caused by an intrinsic stenosis of the ureteric orifice. The position of its orifice may be normal or ectopic.

(2) The sphincteric ureterocele. The orifice is ectopic and lies on the posterior wall of the urethra in the internal sphincter zone. The continuous squeezing action of the internal sphincter during the resting phases of bladder function is the obstructing mechanism in this type. All the ureteroceles in this series occurred on the ectopic ureter of a double ureter anomaly.

Combinations of the stenotic with the sphincteric types may occur though none was seen in this series of cases.

Case Material

The 16 patients in this series exhibited 18 ureteroceles. Eight of the ureteroceles were stenotic and 10 were sphincteric.

The ages of the patients ranged between the newborn period and twelve years; 12 were under two years.

Twelve of the 16 patients were female. In 13 patients the ureterocele was associated with double ureter anomalies (Table 1).

Pathological Anatomy

The stenotic ureterocele

This ureterocele is usually located on the lateral aspect of the base of the bladder. The ureteric orifice lies on the dome of the globular protrusion. All grades of stenosis occur from almost normal to pin-point calibre. In one specimen, the stenosis was shown microscopically to include the vesical mucosa and the adjacent terminal end of the ureterocele. Muscle surrounded in part the narrow segment.

The less stenosed orifice is accompanied by intermittent distension of the ureterocele with each peristaltic wave. The wave passes off with an ejection of a strong spurt of urine from the orifice. The ureterocele then becomes invisible.

Severe stenosis is accompanied by a permanent distension of the ureterocele. The dribble or wetting of urine from the orifice can be visualized at cystostomy. The leakage may increase intermittently to a weak needle-thickness jet. The orifice is too small to be probed with a ureteric catheter. Often it is too small to be seen. It may be set in a smooth transparent bleb-like nipple of mucosa which pouts beyond the oval contour of the ureterocele.

The orifice is situated posteriorly. It is in the internal sphincter zone of the urethra and is invisible from within the bladder. It lies beyond the globular ureterocele.

Its orifice is not obstructive; it may be larger than normal, readily admitting the beak of the urethroscope: but it is completely blocked off by the compression of the internal sphincter muscle of the urethra when the bladder is inactive. It is completely

TABLE 1

DETAILS OF 18 URETEROCELES IN 16 PATIENTS

Stenotic type 8	2	Bilateral and situated on normally placed orifices	2 Right	2 Left	2 Male
	1	Unilateral on normally placed orifice		1 Left	1 Male
	1	Unilateral on the normally placed orifice of conjoined double ureter		1 Left	1 Female
	2	Unilateral on ectopic orifices of double ureters	2 Right		2 Female
Sphincteric type 10	10	Unilateral situated on ectopic orifices of double ureters	8 Right	2 Left	1 Male 9 Female

The size of the ureterocele is determined partly by the degree of stenosis, and partly by the length of the ureter in the submucous layer of the base of the bladder. The ureterocele of the longer ectopic ureter is consequently larger than that of an orthotopic ureter.

The ureterocele has the power of muscular contraction which is co-ordinated to form peristaltic waves.

The microscopic appearances confirm the presence of a muscular wall of the ureterocele, continuous with but thinner than the ureter above and made up of smaller muscle bundles.

The sphincteric ureterocele

Here the ureter has a long and sometimes tortuous course in the submucous layer of the bladder from the intramural tunnel to the internal sphincter zone of the urethra. This form of ureterocele is usually large. Sometimes it nearly fills the bladder. It is centrally situated in contrast to the lateral position of the stenotic ureterocele.

unblocked during micturition when the sphincter is relaxed. Ericsson (1954) made this observation though he regarded the chances of emptying of the ureterocele as limited.

The ureterocele enlarges like a bladder to retain the urine which is excreted by the corresponding kidney. When voiding starts the ureterocele contracts, expels its contents along the urethra and becomes invisible beneath the trigonal mucous membrane of the base of the bladder.

The distension of the ureterocele may be of a degree sufficient to cause pain. Relief of pain is achieved by voluntary micturition.

It is remarkable that the squeeze of the internal sphincter during the filling phase of bladder function on this type of ectopic orifice is so effective that it neither allows leakage proximally into the bladder nor distally to cause wetting incontinence (Fig. IIIa and b). On the other hand, an ectopic orifice opening more distally in the urethra is incompletely obstructed, causes wetting and its

thickened ureter is not visible in the bladder as a ureterocele.

The orifice of the ectopic ureter is usually continent, but in two of the ten cases studied by us both efflux and reflux occurred during micturition so that partial refilling took place at the end of micturition.

This refilled ureterocele forms a bolus which may then be expelled by the detrusor muscle of the bladder as a prolapse down the urethra.

Ureteroceles with both competent and incompetent types of orifice showed active peristaltic contractility when the obstructive internal sphincter was unblocked by the cystoscope.

The excised ureteroceles on microscopy have all shown smooth muscle in the walls.

EFFECTS OF URETEROCELE ON URETHRA AND URETER

The ureterocele may produce secondary obstructive effects on urethra and ureter (Gross and Clatworthy, 1950).

Obstruction of urethra

The ureterocele of stenotic type, when large and tense, may prolapse through or lie over the internal urethral orifice and may partly obstruct the urethra (Campbell, 1951). This, however, was not observed in the three children in this series.

The sphincteric ureterocele is large and often may nearly fill the bladder. It lies over the internal urethral orifice only when the ureterocele is distended. But synchronously with relaxation of the internal sphincter at the beginning of micturition, the orifice of the ureterocele is unblocked. Both bladder and ureterocele then empty together without obstruction to either. Occasionally the redundant vesical mucosa is forcibly evaginated postero-laterally into the space previously occupied by the distended ureterocele.

The urethra is free of obstruction though false radiographic conclusions regarding obstruction may be drawn from cysto-urethrographic appearances in the early phase of micturition. A misleading situation arises when junction is effected between the translucent stream from the ureterocele and the iodide column of the urethra; at this point the radiographic picture may be wrongly interpreted as that of an obstructive filling defect.

This concept is borne out in this series by the unimpaired state of the contralateral ureter.

Prolapse of the ureterocele temporarily obstructs the urethra. This is more likely to

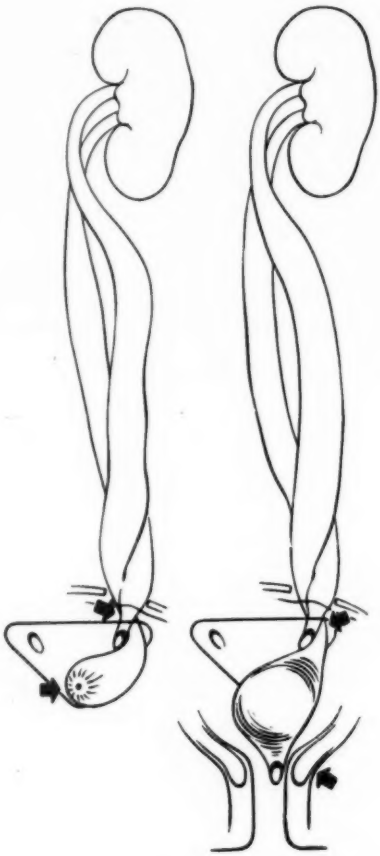


FIG. I. Stenotic and sphincteric ureteroceles (left and right respectively) situated on ectopic ureters. The lower arrows indicate the sites of the obstructions which cause the ureteroceles. The upper arrows show the points at which the turgid ectopic ureters and ureteroceles obstruct the orthotopic ureters and their orifices.

occur in the sphincteric ureterocele which itself encroaches on the urethra. Reduction of the prolapse relieves the obstruction.

Obstruction of ipsilateral orthotopic ureter

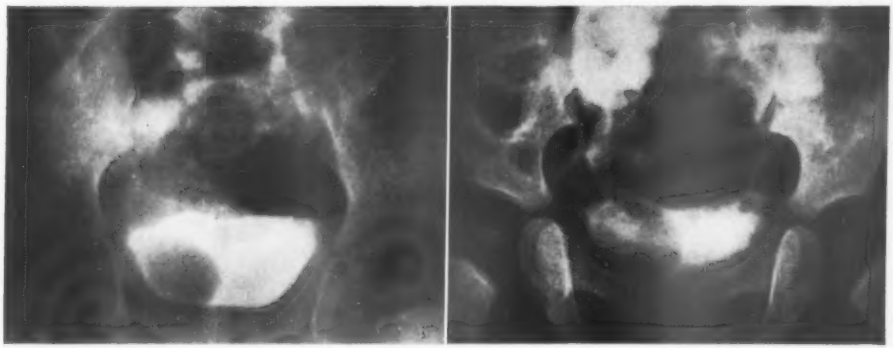
The actual course of the ectopic ureter within the walls of the bladder conduces to obstruction of the orthotopic ureter.

The ectopic ureter traverses the muscular wall of the bladder through a tunnel in common with the orthotopic ureter, then crosses posteriorly to the orthotopic orifice lying in the submucous plane between this orifice and the muscle wall of the bladder (Fig. I).

DIAGNOSIS

The type and special properties of the ureterocele are elucidated in the course of comprehensive routine urinary tract investigation. This includes urine examinations, micturition cysto-urethrography, intravenous pyelography, retrograde cysto-urethroscopy and retrograde pyelography, and tomography when indicated.

The image intensifier has been of great value in assessing the dynamics and emptying of ureteroceles.



(a) (b)

FIG. II. Shows the laterally placed filling defect of stenotic ureterocele unchanged before (a), and after micturition (b). (Reflux has occurred into the ipsilateral orthotopic megaureter in this micturition cysto-urethrographic series.)

The ectopic ureter and ureterocele, when turgid, compress the orthotopic orifice (Fig. I). The orifice may even be stretched with resulting partial impairment of the ureterovesical valve mechanism (Higgins *et alii*, 1951). The ultimate effects are partial obstruction of the ureter and limited reflux, both of which recover after elimination of the obstruction of the ureterocele.

Acquired vesico-ureteral reflux affecting the orthotopic orifice must not be confused with the innate impairment of the valve mechanism which is the basis of the ureteral reflux so commonly associated with the orthotopic orifice in double ureter disease (Ericsson, 1954; Stephens, 1956). This acquired impairment can be expected to recover after appropriate treatment of the ureterocele (3 cases) but the congenital impairment remains permanently (4 cases).

Diagnostic criteria

Typical findings in routine investigations serve to establish the presence of the ureterocele. These are:

(1) On cystography a filling defect in the base of the bladder.

(2) On cystoscopy a rounded protrusion which may obscure the landmarks of the trigone. Sometimes this cyst-like structure discharges its content. Then its walls writhe with peristaltic contractions and its shape dramatically assumes a tortuosity in the base of the bladder; finally it subsides from view beneath the otherwise normal trigonal platform.

But it is the dynamic characteristics of each type which dictate the policy of surgical treatment of the ureterocele. Both the type

and emptying properties are revealed by micturition cysto-urethrography during and at the end of voiding when the bladder is well night empty (Figs. II and III).

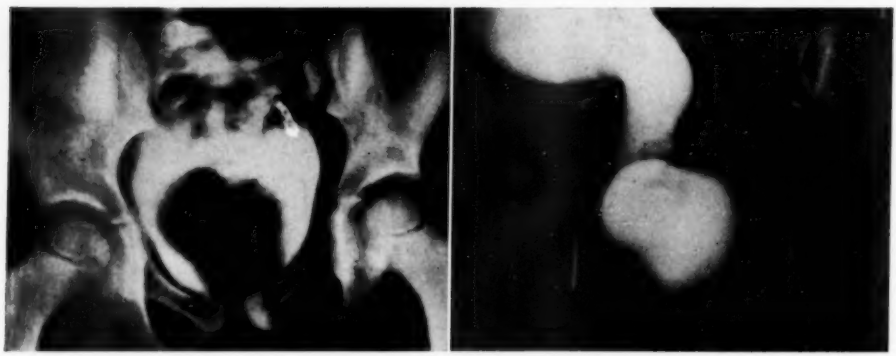
The features that have discriminatory value are: a globular "filling defect," which is obvious (outlined by a shadow of iodide remaining in the bladder) during and at the end of micturition, and which indicates the stenotic type of ureterocele (Fig. II); a ureterocele which is filled with iodide at the end of micturition, the sphincteric type, its orifice being incompetent and permitting reflux from the urethra during micturition (Fig. III); and lastly, that which vanishes during micturition and becomes invisible at the end of micturition, a condition which is most likely to be the sphincteric type with a competent ureteric orifice.

patient and the presence of associated abnormalities of the other ureters and their corresponding kidneys may influence a decision between conservative and radical methods of treatment.

When renal function is worth preserving

In the case of the stenotic variety, the target of surgery is the ureterocele and its obstructing mechanism. Gross and Clatworthy (1950) recommend for infants and children a minimal division of the ureterocele roof sufficient to relieve obstruction and yet not so large as to cause reflux. They advise the suprapubic route in small patients.

In three children in this series, vesico-ureteral reflux followed partial resection of the roof. Multiple micturition (Stephens, 1957) now controls the reflux pooling of



(a) (b)

FIG. III. Shows the medially situated filling defect of the sphincteric ureterocele before micturition (a) and after micturition (b), when the ureterocele is evident because of its contained iodide, indicating that its orifice is incompetent.

Treatment of the ureterocele depends in large measure on its emptying ability, as demonstrated radiographically, but it is also influenced by the function of the corresponding kidney. Routine tests, especially the intravenous pyelogram, are used to determine its function.

TREATMENT OF URETEROCELE

Sufficient information has now been obtained to deal not only with the ureterocele but also with the corresponding kidney. Treatment of the ureterocele depends mainly on the relative value of the kidney to the patient. Other factors such as the age of the

urine in two, but the third child died in the very early post-operative period from proteus septicaemia.

In the sphincteric ureterocele, in which the normal or large calibre orifice lies in the zone of the internal sphincter, the ureterocele must be partially de-roofed proximally from the orifice in order that it may freely open into the bladder. This will circumvent the obstructive mechanism. Transurethral resection or suprapubic procedures are both satisfactory methods of achieving the objective.

In 2 patients in this series, the ureterocele was sparingly de-roofed by transurethral resection. Follow up studies show that the uretero-vesical valve mechanism is preserved. In 5 others free reflux followed the resection operation which in some was more radical.

Improvement of renal function follows, but vesico-ureteral urinary reflux with pooling and stagnation may occur.

Multiple micturition therapy has again eliminated infection caused by reflux pooling of urine.

In one infant, both the ureterocele and the double kidney with its very large megaureters were excised to combat infection.

When it is a case of the sphincteric ureterocele nephrectomy and partial ureterectomy are adequate for the non-reflux type. Here the ureterocele is unobstructed during micturition. After nephrectomy it empties itself and does not refill by reflux. Infection in the ureterocele is unlikely to persist or recur.

In the case of the reflux type, in addition to nephrectomy and ureterectomy, the ureterocele itself should be de-roofed or excised

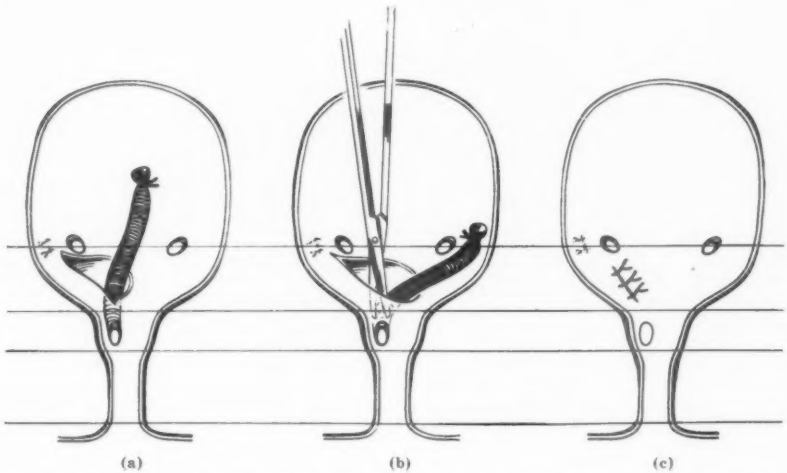


FIG. IV. To show the steps in the total excision of the lower ureter and the sphincteric ureterocele.

(a) The ureter has been dissected and passed through the intramural tunnel into the incision in the trigone.

(b) The distal end of the ureter is then excised in the submucosal plane and detached from its orifice.

(c) The trigonal incision is then sutured.

(The horizontal lines divide the bladder and urethra into three zones occupied by the trigone, internal sphincter, and the external sphincter respectively.)

When renal function is not worth saving

In the case of the stenotic ureterocele nephrectomy (or hemi-nephrectomy) and excision of the greater part of the ureter should be adequate to cure this condition. The ureterocele slowly empties itself through its narrowed orifice and does not refill. Persisting infection in this type is unlikely.

If, however, infection in this ureteric stump occurs, excision of the collapsed ureterocele and ureteric stump is necessary at a later date.

completely for the following reasons: The ureterocele will refill during each act of micturition and remain as a globular enlargement, distended by newly imprisoned refluxed urine. This predisposes to infection and to prolapse. In addition, the distended ureterocele acts as a partial obstruction to the orthotopic ureter. The orthotopic orifice is compressed between the trigone and the distended ureterocele (Fig. I).

Total excision of the sphincteric ureterocele was performed in two children with double ureters (Fig. IV).

The lower end of the corresponding ureter was dissected in the extraperitoneal plane, whilst the ureterocele was isolated in its submucous plane through a short incision on the trigone. The ectopic ureter was then passed along its intramural vesical tunnel into the incision in the trigone. With a little further freeing of the ureterocele itself, the excision was completed. The mucosal incision in the bladder base was then oversewn and the ectopic orifice in the urethra was left to heal over spontaneously.

This procedure has been applied to 2 children with ureterocele and in addition to 3 others with ectopic urethral ureters. Of these the orthotopic ureter has been conserved in three; post-operative vesico-ureteral reflux occurred in one of these orthotopic ureters necessitating multiple micturition therapy, but the other two have maintained normal ureterovesical continence.

RESULTS OF TREATMENT

The results of treatment of the stenotic type ureteroceles have already been discussed.

It now remains to correlate the treatment of the 10 patients exhibiting the sphincteric type of ureterocele.

Two are now in good health after partial resection of the ureterocele by the transurethral route. The corresponding ureteric orifice is now competent.

In 4 others the resection was followed by reflux. Three are now well but require multiple micturition therapy. One infant required further hemi-nephrectomy operation for uncontrollable infection and is now cured.

Nephrectomy or ureteric ligation operations with or without excision of the ureteroceles were performed on the remaining four children. Three are now in good health, apparently cured, but the fourth, an infant of 15 months, died four months after a hemi-nephrectomy operation from causes unknown.

CONCLUSIONS

Ureteroceles are classified in this paper into two groups, stenotic and sphincteric, depending on the cause of the obstruction — stenotic, when its orifice is structurally narrow, and sphincteric when the ectopic ureteric orifice is obstructed by the grip of the internal sphincteric muscle of the urethra.

Their dynamic properties, demonstrable by micturition cysto-urethrography, give the cue to diagnosis and provide a logical basis for treatment. The stenotic ureterocele, in which the stenosis is severe, remains apparent throughout as a filling defect against the iodide content of the bladder. The sphincteric type in which the ureterovesical valve mechanism is competent vanishes during micturition when the grip of the internal sphincter relaxes; when its valve is incompetent, the filling defect caused by the ureterocele disappears during micturition only to reappear filled with iodide at the end of micturition.

Surgical treatment depends primarily on the overall renal function and that of the kidney corresponding to the ureterocele.

When the renal function in the corresponding kidney is important to the patient, operation on the ureterocele is performed to circumvent the obstruction. If vesico-ureteral reflux persists after operation, chemotherapy and multiple micturition are required to remedy infection.

When the corresponding kidney function is worthless to the patient operative removal of the defunct kidney tissue and the ureter will suffice provided the ureterocele empties itself subsequently and does not permit pooling of urine by reflux. On this reasoning, both the stenotic ureterocele and the sphincteric type with the competent ureteral valve mechanism are unlikely to require additional treatment. On the other hand, the sphincteric ureterocele with an incompetent ureteral valve requires excision or partial resection. The ureterocele, tense with imprisoned urine, would predispose to prolapse and infection, and would partially obstruct the orthotopic ureter as well.

Multiple micturition techniques have now successfully overcome the bogey of post-operative ureteric reflux and its accompanying pyuria in children. In the infants too young to co-operate, intense chemotherapy is necessary until such time as prophylactic multiple micturition therapy becomes possible. A more conservative attitude in the management of ureterocele can be adopted now that infection caused by vesico-ureteral reflux can be overcome by this simple technique.

ACKNOWLEDGEMENTS

My thanks are due to the many members of the Senior Medical Staff who have referred these patients to me for investigation and treatment.

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RECURRENT ACUTE PANCREATITIS

By Terence W. Horne

Hobart

RECURRENT pancreatitis may be defined as that condition in which the patient suffers from recurrent attacks of acute pancreatitis with more or less complete remission of symptoms between attacks. Sometimes the attacks may occur so closely together that the patient is almost continually in pain. The pathology of these attacks is usually that of acute oedematous pancreatitis but at times acute haemorrhagic pancreatitis may occur and this may be fatal.

The condition of recurrent pancreatitis has been investigated clinically at the Royal Hobart Hospital for approximately the last three years.

Diagnosis

In severe cases if the presence of the disease is not suspected laparotomy is usually performed and the diagnosis made by inspection. Milder cases such as occur in recurrent pancreatitis are not usually subjected to immediate operation and here misdiagnosis is common. The commonest misdiagnosis is probably that of acute cholecystitis and, indeed, the symptoms and signs of the two conditions may be identical. Other common errors are to diagnose "leaking peptic ulcer," chronic peptic ulcer, acute appendicitis and intestinal obstruction. In our experience the clinical picture of recurrent pancreatitis may mimic that of all the above conditions; so much so that acute pancreatitis may be called the "masquerader of the abdomen."

The frequency with which a correct diagnosis of acute pancreatitis is made is directly proportional to the zeal displayed in looking for the condition. Thus before 1953 only 2 to 4 cases per year are filed in the records of the Royal Hobart Hospital whereas subsequent to 1953 the recorded incidence of the disease has approximated to that of ruptured peptic ulcer. This increase has been due to greater awareness of the disease and to the routine use of the serum amylase estimation.

Clinical features which point to a diagnosis of acute pancreatitis are as follows:—

1. Pain is often continuous and lasts for several hours or days at a time. This serves to distinguish it from biliary colic which usually lasts only two to three hours, but not from the pain in acute cholecystitis. Sometimes the pain is of short duration and occurs immediately after the ingestion of food, mimicking the pain of peptic ulcer. We have not found the patient's description of the pain nor the site of the pain to be of any value in differential diagnosis.

2. The site of tenderness is helpful. It is difficult to distinguish with any measure of reliability between tenderness of the head of the pancreas and tenderness of the gall-bladder, but tenderness of the tail of the pancreas when present, is a reliable guide to the diagnosis. Thus, one patient referred because of gall-stones gave a history of symptoms resembling those of peptic ulcer. Tenderness of the tail of the pancreas excited the suspicion of pancreatitis and subsequent investigation showed this to be the cause of her pain.

The serum amylase estimation gives by far the most valuable help in the diagnosis of recurrent pancreatitis. We consider it is obligatory to determine the serum amylase in all cases of abdominal pain where the diagnosis is not certain, and, in particular, in cases diagnosed as acute cholecystitis. The interpretation of the serum amylase value is by no means straightforward and we have investigated this aspect.

Burnett and Ness (1955) determined the serum amylase in a consecutive series of 350 cases admitted to hospital as abdominal emergencies. They found consistently high values (greater than 1,000 Somogyi units) in acute

pancreatitis; they also found high values in two cases of perforated peptic ulcer and two cases of intestinal obstruction with strangulation.

We have confirmed these results on a smaller scale. In 50 cases whose main symptoms was abdominal pain we did not find one with a serum amylase greater than 400 Somogyi units other than those with acute pancreatitis, perforated peptic ulcer or intestinal obstruction with strangulation. One man with a perforated duodenal ulcer of seven hours duration had a serum amylase of 1,600 Somogyi units. Another man with a small bowel obstruction with strangulation, of 48 hours duration, had a serum amylase of 800 Somogyi units. We have found values up to 400 Somogyi units in a variety of conditions including acute cholecystitis. Our present assessment of the situation is that with a serum amylase of 600-800 units we feel that pancreatitis is a strong possibility and with values of a 1,000 units or more the diagnosis is definite, providing always that perforated peptic ulcer and intestinal obstruction with strangulation can be confidently excluded.

This latter consideration implies that in many cases of severe acute pancreatitis operation will still be performed. A plain X-ray of the abdomen shows gas under the diaphragm in something like 70 per cent. of cases of perforated ulcer. Absence of free gas is, therefore, by no means reliable evidence against the presence of a perforation. Consequently the surgeon's clinical judgment remains the most important factor in the decision, and when there is doubt it will be usually safer to operate.

There are three other points to be considered when interpreting the serum amylase value.

These are:—

1. The time between the onset of the pain and the taking of the blood.
2. The time between the taking of the blood and the performance of the estimation.
3. The significance of the normal value.

Our results confirm that the serum amylase takes several hours to reach a high level and tends to fall to within normal limits after three or four days. The practical significance of this is, of course, that a normal serum amylase may be without significance if the blood is taken too soon or too late. A sample taken, say, two hours after the onset of the pain is unlikely to show a significant elevation of the amylase, whereas another sample taken twelve hours later may give a diagnostic value. A serum amylase estimation should, therefore, be interpreted with due regard to the time factor.

Observations on a small number of cases have shown that the amylase activity of blood stored at room temperature does not deteriorate over a twelve-hour period. Thus it should be possible to have the test done on patients living in areas remote from biochemical facilities. Also in cases which are admitted at night with pain of say three days duration, blood can be taken at a time when the amylase is more likely to be still high and examined at leisure the next day.

The significance of the normal serum amylase has already been discussed with regard to the time factor. We have found that in some cases of proven pancreatitis the amylase may be normal in one attack and raised in another even when the blood has been taken at the optimum time of twelve to twenty-four hours after the onset of pain on each occasion. This means that when one suspects the diagnosis of pancreatitis on clinical grounds, one should not be put off by one or two normal serum amylase values but should repeat the test several times if necessary.

TREATMENT

The treatment of recurrent pancreatitis is not very satisfactory. We have found that in some cases there is a definite relationship between the occurrence of attacks and the excessive ingestion of fat or alcohol. These cases are usually kept free from pain by limiting the intake of the offending substance. More often, however, no such relationship can be established.

Many surgical procedures designed to relieve this condition have been described. Stimulated by the work of Starr of Sydney (Starr, 1954) we elected to try the operation of sphincterotomy, i.e. division of the sphincter of Oddi.

The rest of this paper is based on a study of fifteen cases where sphincterotomy was performed. The first operation was performed in July, 1954, and the last in November, 1956, so that the period of follow-up varies from nearly three years to six months at the time of writing.

The indications for the operations are given in Table 1.

TABLE 1

Indication	*No. of Cases*
Recurrent pancreatitis resistant to medical treatment	13
Persistent pancreatic fistula following drainage of a pancreatic pseudocyst in a patient with recurrent pancreatitis	1
Obstructive jaundice	1
Total:	15

The patient with obstructive jaundice is included in this study because of the unusual operation findings and their possible relationship to the aetiology of recurrent pancreatitis.

This female, aged 61, presented in June, 1955, with jaundice of two weeks duration. The illness had been painless except for a transitory mild pain under the left costal margin at the onset. There was a past history of flatulent dyspepsia for none years.

At operation on 15th July, 1955, the gall-bladder contained cholestrol stones and was removed. The common duct was found to be dilated and was explored. No stones were found and a sound passed down the duct appeared to enter the duodenum freely. The abdomen was then closed with T-tube drainage of the duct.

Her post-operative course was unsatisfactory. It became obvious that no bile was entering the duodenum and a cholangiogram showed a stricture of the lower end of the common duct.

At re-operation on 24th July, the duodenum was opened and the ampulla of Vater found to be thickened and stenosed. A sound could not be passed even under vision. Sphincterotomy was performed and the subsequent convalescence was satisfactory.

There has been no indigestion since.

There are two main points of interest here:—

1. The great difficulty in assessing whether a sound has actually passed through the ampulla into the duodenum. At the first operation the point of the sound appeared to pass right down into the third part of the duodenum, but subsequently under vision it was found that the mucosal papilla was merely being pushed in advance of the sound by virtue of the elasticity and redundancy of the duodenal mucosa. We have since confirmed this observation in several other cases.
2. The aetiology of the obstruction, namely a stricture of the ampulla of Vater. The cause of this is hard to say, but it is important to remember that such a stricture may occur. This point will be elaborated later.

Of the 14 cases where sphincterotomy was performed for pancreatitis, 10 were females and 4 were males.

The ages of the females varied from 21 to 71 years, with an average of 39 years. The ages of the males were 33, 48, 31 and 52 years.

The duration of symptoms prior to operation varied from three months to seventeen years, with an average of three years and three months.

In four cases, the gall-bladder had been previously removed. In two cases gall-stones were reported to have been present, and in two the gall-bladder was said to have been normal. Each of these patients described their pancreatic pain as being identical with that experienced prior to the cholecystectomy. This fact admits of two explanations. Firstly, that pancreatic pain and biliary pain are often indistinguishable; and secondly, that in at least two of the cases, the gall-bladder was removed when the pancreas was the pain-producing viscus. We believe that this is the explanation of many so-called cases of "post-cholecystectomy syndrome." In fact, we believe that, after stone in the common duct, pancreatitis is the commonest cause of recurrent pain after cholecystectomy, whether stones were present or not.

The actual operative procedure carried out was not the same in all patients.

Our original intention was to divide the sphincter of Oddi by a transduodenal approach, expose the interior of the ampulla of Vater, identify the orifice of the pancreatic duct and then to divide the septum between the terminal portions of the pancreatic and common bile duct. This is the operation described by Starr (1954) and is based on his belief that the cause of recurrent pancreatitis is a stricture of the terminal portion of the pancreatic duct.

This was done in our first case, a female of 64, and she obtained an excellent result, being free from pain nearly three years later.

Our second case was a man of 33 presented in October, 1954, with a large pseudocyst of the pancreas. This was drained and a pancreatic fistula resulted which showed no sign of closing after three months. In view of the work of Doubilet and Mulholland (1953) on the use of sphincterotomy in pancreatic cysts and fistulae we decided to try this method rather than those where the fistula is anastomosed to stomach or bowel.

At operation on 10th January, 1955, a phlegmonous pancreatitis was still present and it was not possible to do more than a sphincterotomy. The pancreatic duct was not seen. In spite of this the fistula closed after six weeks and the patient has had no recurrence of his pancreatitis since.

At this stage we became sceptical about the necessity for interfering with the pancreatic duct, especially as anatomical studies (Hughes and Kernutt, 1954) had shown that the diameter of the pancreatic duct in its terminal part was normally of the order 1.00 mm.

We decided to attack the pancreatic duct only when it was easy to find and to compare the results.

In the next 10 cases, the pancreatic duct was dealt with in five. In only one of these was there any suspicion of narrowing of the duct and this was equivocal. In the remainder, lachrymal probes could be passed up the duct with ease.

Comparison of the two groups is difficult because of the small number involved and because in some of these cases additional operative manoeuvres were carried out (choledocho-duodenostomy and cholecystenterostomy in two cases where the thickened pancreas had compressed the bile duct, and cholecystectomy plus choledocholithotomy in another). However, there does not seem to be any real difference in the results in the two groups and we feel that it is unnecessary to interfere with the pancreatic duct.

Another reason why we decided to leave the pancreatic duct alone is that in Cases 12 and 13, troublesome bleeding occurred from the pancreas and this was difficult to control. In Case 12, it was controlled by a T-tube with a long limb lying in the ampulla. This patient developed a severe post-operative pancreatitis, and it was thought that this may have been due to the T-tube occluding the pancreatic duct. Consequently in Case 13, the bleeding was controlled rather inadequately by sutures only. This patient also developed post-operative pancreatitis and this time we postulated a block of the duct by oedema and blood clot.

In Case 14, therefore, the duct was merely probed to establish patency. This patient died three days after operation from what was clinically acute pancreatic necrosis.

In Case 15, sphincterotomy alone was performed, and this patient also delevoped post-operative pancreatitis though fortunately she made an uneventful recovery.

It is difficult to say, therefore, what part, if any, the manipulation of the pancreatic duct played in these unfortunate results. We consider, therefore, that since it does not seem to be necessary and may actually be harmful, it is better to leave the pancreatic duct alone.

To sum up then on the question of operative procedure, if it is decided to use this method of treatment, we recommend the following technique:—

The abdomen is opened through a suitable incision (usually a right upper paramedian, or a transverse incision) and a careful inspection is made. The common bile duct is exposed and opened and a sound is passed

down to the ampulla of Vater. The duodenum is then opened longitudinally or obliquely over the point of the sound. Usually the sound will be found not to have entered the duodenum. The point is then manoeuvred through although sometimes it is necessary to incise the mucosa over the sound. The margins of the ampullary orifice are then secured with stay sutures and the sound withdrawn. The superior wall of the ampulla is then incised for a distance of about a third of an inch. Bleeding is controlled by sutures which are also used to anchor the divided edges back to the duodenal mucosa.

The duodenal wall is then closed transversely in two layers and a T-tube inserted into the common duct.

The gall-bladder is removed if stones are present. Doubilet and Mulholland (1956) recommend removal of the normal bladder on the grounds that it cannot function as a reservoir in the absence of a competent sphincter of Oddi. We have no definite views on this point.

The abdomen is then closed in a routine manner.

Mortality

There were two deaths in this series, a mortality of 13 per cent. The first was Case 12, a man aged 52. He was the man who developed a severe attack of post-operative pancreatitis. Subsequently an abscess in the region of the head of the pancreas was drained and a biliary fistula followed. He also developed liver failure and slowly went downhill to die of pneumonia three months after operation. No autopsy was allowed.

The second death was in Case 14, a young woman of 20. She developed what was clinically a post-operative pancreatic necrosis with severe shock and died three days after operation in spite of heroic attempts to resuscitate her. Unfortunately no autopsy was done.

Both these deaths can be attributed directly to post-operative pancreatitis and this problem will be discussed in more detail later.

Complications

These cases had, of course, their share of the usual complications incident to surgery in the upper abdomen.

In 11 of the cases a T-tube was not used, the common duct being sutured. In five of these a prolonged biliary fistula occurred. This was surprising as it was expected that division of the sphincter of Oddi would facilitate entry of bile into the duodenum. That it did not do so, we attribute to oedema of the cut margins of the ampulla. The above is the reason why we recommend the use of a T-tube.

Another and more serious complication was the occurrence of pancreatitis after operation. This occurred in four cases — the last four in the series. We have already discussed the possible role of operative technique in the production of this complication.

Post-operative pancreatitis is, of course, well known after other operations and not necessarily those on the biliary tract. Thus, in the past two years at least four fatal cases have occurred in Hobart — three after exploration of the common bile duct and one after a partial gastrectomy for chronic duodenal ulcer. In two of the biliary explorations there is some evidence that the patients suffered from pancreatitis prior to operation. The patient with the ulcer had a local pancreatitis from penetration by his ulcer. At operation there was no interference with his pancreas, a Bancroft procedure being carried out.

Post-mortem examination in one of these cases showed the ampulla of Vater had been split by the passage of a sound and was markedly oedematous. There were no thrombi in the larger pancreatic vessels.

Our impression is that post-operative pancreatitis occurs mainly in patients who have previously had attacks of pancreatitis although we have no proof of this. If this is so, it means that instrumentation of the lower end of the common bile duct is a dangerous and potentially lethal procedure in such patients, and the greatest care should be taken to ascertain whether patients with gall-stones do not also have pancreatitis before such procedures are carried out.

RESULTS

Excluding the 2 cases who died and the case which was operated on for obstructive jaundice, the results of operation in the remaining 12 cases are given in Table 2.

TABLE 2

RESULTS OF OPERATION

Good result — no further attacks of pain	8
Fair result — some further pain for a few months only	3
Poor result — still getting pain eighteen months after operation	1

The patients who had fair results were those who had either a long history (seventeen years in one) or some degree of permanent damage to the pancreas (marked fat necrosis and fibrosis). Although these patients have had further attacks of pancreatitis since operation, these attacks have tended to become less frequent and it seems probable that the eventual outcome will be satisfactory.

The patient with the poor result had neither a long history (five months) nor marked change in the pancreas (slight thickening only). We have no explanation of this failure. However, she asserts that her attacks are less severe and does not resent the operation.

These results indicate that providing the patient survives the operation, that his or her symptoms will be cured or at least relieved in most cases. It is the bugbear of post-operative pancreatitis which spoils the results and makes us rather hesitant to advise the operation in the future.

DISCUSSION

The exact aetiology of recurrent pancreatitis remains obscure. Doubilet and Mulholland (1956) believe that the cause is primarily due to obstruction at the sphincter of Oddi. This obstruction may be due to spasm or to an actual fibrotic stenosis. They believe that spasm is the usual cause and that it may be emotional in origin.

Our experience supports this. We have found what appeared to be a fibrous stricture of the ampulla of Vater in two cases, and in one of these actual obstructive jaundice was present. We have tried on several occasions to study the wall of the ampulla histologically but this has been unsatisfactory for technical reasons. Nevertheless, in most cases the ampulla has appeared normal macroscopically, and the obstruction at this site presumably spasmodic in origin. Further study of this point is needed.

We have been impressed by the fact that most of the individuals in this series have been very nervous and even neurotic, and in some a relationship between the onset of the disease and some psychological trauma is evident. Thus, several patients dated their symptoms from a pregnancy. One patient suffered also with migraine; one, having been cured of her pancreatitis, developed mucous colitis, and another had an acute neurotic disturbance during her convalescence. A man of 36, not in this series, suffers with both migraine and recurrent pancreatitis and gets attacks only when confronted with some difficult problem in connection with his work as a draughtsman.

We have wondered whether post-operative pancreatitis may not be a response to the acute mental trauma of a major operation. It seems certain that some cases are due to oedema of the ampulla consequent upon operative trauma. Perhaps it is a combination of the two factors which is so disastrous.

In any case, it is clear that operations on the lower end of the common bile duct, including sphincterotomy, are accompanied by an unsatisfactory morbidity and mortality. Some safer procedure to reduce the resistance at the sphincter of Oddi is required. Clearly, sphincterotomy is mandatory to deal with a fibrous stricture, but spasm is said to be abolished by vagotomy. Recently Mallet-Guy and Rose (1956) have described a method to distinguish spasm from stricture, at operation. With this method it may be possible to treat the majority of these patients by vagotomy and to reserve sphincterotomy for cases where a stricture is present. We hope to investigate this possibility.

SUMMARY

1. Acute pancreatitis is not uncommon and may masquerade as many other intra-abdominal lesions.
2. The serum amylase estimation is of the greatest value in diagnosing acute pancreatitis, although it does not replace clinical judgment.
3. Sphincterotomy of the sphincter of Oddi in the treatment of recurrent pancreatitis gives satisfactory end-results but carries a high morbidity and mortality, and in this respect it is an unsatisfactory operation.
4. The possible aetiology of recurrent pancreatitis is discussed and evidence is given that this may be obstruction at the ampulla of Vater due to either a fibrous stricture or more commonly a spasm of the sphincter of Oddi.

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THE CONTROL OF BLEEDING IN EXTRADURAL HAEMORRHAGE

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AN extradural haemorrhage is an acute surgical emergency which may have to be treated by a surgeon who is not familiar with all the methods of controlling intracranial bleeding, nor may he have all the tools available for such techniques. The classical extradural haemorrhage comes from the middle meningeal vessels and fills the middle fossa. However in about a third of cases the bleeding arises from an intracranial venous sinus (Rowbotham, 1945) and in a few no major bleeding point can be identified. The amount of the blood clot is increased as the haematoma separates the dura from the skull and starts oozing from small vessels. There is soon a snowball effect as the intracranial pressure rises and hastens the rate of bleeding. The onset of coma and an obstructed airway are the terminal events in the unrelieved vicious spiral.

The treatment of the extradural haemorrhage in the middle fossa has become standardized (Jefferson, 1956). A subtemporal burr hole is made which is enlarged with bone nibblers and the bleeding vessels are sealed by endotherm coagulation. When the haemorrhage is at other sites further burr holes are made. It is in these cases that the bleeding may come from a venous sinus and where the advent of the absorbable haemostatics has been such a great advance. Many of the standard texts indicate that this is all there is to successful treatment. The high mortality of the condition being due to delay in operating and to associated brain damage rather than to any failure to allay the bleeding. Other surgeons have not been so fortunate and the control of the bleeding may be very difficult and recurrences are reported. In the Department of Neurosurgery, Dunedin Hospital, there have been 16 cases of extradural haemorrhage treated surgically and 5 died. There was an appreciable re-collection of clot which partly contributed to the fatal issue in 2 of these 5 cases. In another patient the wound had to be packed with gauze before the bleeding could be arrested. Two personal cases are now reported where considerable difficulties in haemostasis were encountered.

Case 1

M.B., a 33-year-old married woman, was admitted to Dunedin Hospital on 25th July, 1951, at 9.15 a.m., soon after having been knocked off her bicycle. She was conscious but amnesic for the accident. There was bleeding from the left ear but there were no other neurological features. At 5.30 p.m. she became hard to rouse. The pupils were small, equal and reacted to light. The plantar responses were extensor. The pulse chart showed a progressive slowing from 80 to 56 over the day. Shortly, the left pupil became dilated and there was an episode of decerebrate rigidity.

Operation. A left temporal burr hole was made under local anaesthesia. A huge extradural clot was removed. The burr hole was enlarged and the torn middle meningeal artery was diathermied close to the foramen spinosum. There continued to be a diffuse vascular oozing from the dura and many of the bleeding points were inaccessible owing to the posterior extent of the dural stripping. By now she had become very restless.

The skin incision was closed loosely. The whole of the head was shaved and a general anaesthetic was given, a large temporo-parietal bone flap was raised. The many bleeding points were coagulated with the endotherm machine. The suturing of the dura up to the pericranium finally produced a dry field. The wound was not drained. Inspection had shown that a fissure fracture ran across the floor of the middle fossa into the petrous part of the temporal bone.

The patient regained consciousness five hours after the operation. She made a good recovery and left hospital on 8th August, 1951.

Comment. Her restlessness made a general anaesthetic imperative for the extended procedure of an osteoplastic flap which was the only way of getting at the vascular weeping from the dura.

The use of an osteoplastic bone flap in extradural haemorrhage has been a matter of controversy. Gurdjian and Webster (1942)

credited Krause with originating the method in 1905. In recent years Raaf (1948), Robertson (1952), Schneider (1955) and Jefferson (1956) said that an osteoplastic flap might be needed when the haematoma was extensive or when other haemostatic measures had failed. King (1943) and Munro (1952) considered that the method was unnecessary.

Difficulties in arresting the diffuse vascular oozing from the dura certainly exist and Gurdjian and Webster (1942) in their review mention the use of such methods as packing the extradural space with gauze, ligature of the external carotid artery and drainage with a rubber tube. Although King condemned the use of a flap he did have to use packing and ligature of the external carotid artery. McKenzie (1938) was troubled by persistent oozing from the dura and recommended gauze packing or the making of further burr holes and suturing the dura up to the scalp and leaving the wound open. Lewin (1949) drained half of his cases but only used an osteoplastic flap once in his 26 cases. James and Turner (1951) stated that it might be necessary to stitch the dura up to the temporal fascia to control the venous oozing. Bradley (1952) and Schneider (1955) made similar suggestions. Botterell (1952) recommended gauze packing and sewing the wound open when there was weeping from a vascular dura.

Many of these difficulties are due to a failure to close the dead space left by the evacuated haematoma. Usually once the clot has been removed and the major vessel controlled, the brain pushes the dura out into contact with the skull and the capillary ooze is stopped. McKenzie noted that this failure of the brain to expand was commonest in delayed cases. Gauze packing is hardly an adequate substitute for an extradural haematoma when persistence of symptoms may be due to an enlarged extradural space. The use of an osteoplastic flap has the merit that it allows haemostasis of the exposed dura. The dead space is then reduced by stitching the dura up to the surrounding tissues (Poppen, 1935). The disadvantages of a flap are that specialized instruments are needed and the state of the patient may make the procedure difficult even for the experienced. Often the patient becomes wildly restless after removing the clot under local anaesthesia. Turning a flap under these circumstances is hazardous and the surgeon will be wise to resort to general anaesthesia, if available. Recently we have used another method which is simple and promises to do away with these difficulties.

Case 2

J.M.O., a boy aged 14 years, was admitted to Dunedin Hospital on 8th August, 1956. At 5 p.m. the evening before he had been kicked on the head but had not been made unconscious. He went home and vomited throughout the night. The next morning he went to sleep and by midday his mother could not rouse him. He was admitted to hospital in the late afternoon in a semicomatose condition. The left pupil was dilated and there was a right-sided weakness with increased tendon reflexes and an extensor plantar response on that side. There was bruising of the left temple. Radiographs of the skull were normal.

Operation. Under local anaesthesia a left temporal burr hole was made and a large extradural haematoma was found which filled the middle fossa from the sphenoid ridge to the occiput. The burr hole was enlarged but no definitive bleeding vessel could be found. Numerous bleeding points were sealed with the diathermy. At closure the operative field was dry but the dura had not come out particularly well. A drain was not used. As soon as the clot had been turned out he regained consciousness.

Course. Over the next four hours he remained alert and the pupils were equal. Then he became comatose with a dilated left pupil and bilateral extensor plantar responses. The wound was opened with him lying on his right side. There was a massive recurrence of the extradural haematoma. A more extensive bone removal was done with bone nibblers. Despite diathermy and the generous use of gelfoam the dura persistently oozed. Matters were made worse by the patient becoming abusively obstreperous. Division of the middle meningeal artery at the foramen spinosum made no difference to the bleeding. A small nick was made in the dura and Ringer's solution was injected into the subdural space. The dura showed no signs of staying expanded and the incision was closed with a stitch.

A lumbar puncture was done and he was already suitably placed for this. Forty c.c. of Ringer's solution were injected into the subarachnoid space. The dura came out into tight contact with the skull and the bleeding ceased at once. The wound was closed over a soft rubber drain. He made a rapid recovery and the drain was taken out on the next day but there was no blood on the dressing.

Comment. This recurrence of the haematoma was due to the extensive dural separation from the skull and the failure of the brain to expand after the clot had been

removed. Owing to his maniacal state it would have been impossible to have turned a flap under local anaesthesia. We were gratified at the rapidity with which the subarachnoid injection stopped the bleeding. The drain served no useful purpose. It is probable that similar treatment would have been effective at the first operation and also in Case 1. This is an uncommon case of extradural haematoma as there was no fracture of the skull and the major vessels were intact.

Recurrence of an extradural haematoma has received some mention in the literature but it is hard to assess its incidence. McKenzie (1938) had one case out of his 20 but the patient also had a ruptured liver at autopsy. Ellis (1938) reported a successfully treated case. Munro and Maltby (1941) in 44 cases had one death from recurrence of the haematoma, but an unstated number occurred and were treated successfully. Reichert and Morrissey (1941) reported two cases of recurrence due to persistent venous bleeding. Sartorious and Humphries (1946) had one recurrence with recovery in 20 patients. Raaf (1948) had four recurrences in the 20 patients which he operated upon. Lewin (1949) had three cases of recurrent bleeding in 26 patients and although all were recognized only one survived further surgery.

LaLonde and Gardner (1948) and Robinson (1955) have shown that the results of treating chronic subdural haematomas are improved by injecting normal saline or Ringer's solution into the subarachnoid space after the clot had been removed. The method is effective because it expands the compressed brain and closes the dead space, reduces the tentorial herniation and counteracts any intracranial hypotension. McElwee and Ray (1950) used the technique with success to stop the venous bleeding from a case of acute subdural haematoma when they could not get at the bleeding vessel. Gillingham (1954) routinely injected 100 c.c. of normal saline into the subarachnoid space after the removal of a traumatic intracranial clot. This was to reduce the tentorial herniation and no haemostatic properties were claimed for the manoeuvre. The use of this method in Case 2 was a success and is worthy of extended use.

It is suggested that in an extradural haemorrhage where the dural oozing persists after the control of the major vessel or when the dead space is not obliterated by the expansion of the brain then enough normal saline solution should be injected into the lumbar subarachnoid space to overcome these difficulties. It is unlikely that more than 100 c.c. will be needed and at the worst no harm is done. Only in failure of this simple method should the use of an osteoplastic flap be considered with its implication of a general anaesthetic. There is no place for gauze packing or ligature of the external carotid artery in these troublesome cases.

SUMMARY

Two cases of extradural haematoma are described where there was difficulty in controlling the bleeding. One case was treated by an osteoplastic bone flap. The other case developed a recurrence of the bleeding and this was arrested by the subarachnoid injection of Ringer's solution. The merits of the subarachnoid injection of fluid in the treatment of intracranial haematomas are stressed.

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THE PATHOLOGY OF FRACTURE IN PAGET'S DISEASE

By MAX E. LAKE

Sydney

OSTEITIS deformans was noted in prehistoric bones by Fisher (1935) and Sugarbaker (1940). In the large literature of Paget's disease since Czerny (1873) and Paget (1876), there has been widespread mention of fracture as one of its principal disabilities. Schmorl (1930, 1932) and Jaffe (1933) have contributed much of our knowledge of the pathology of fracture in Paget's disease and with Braidsford's (1938) observations on radiological aspects of fracture in Paget's disease form a foundation on which to further consider the pathology.

Figs. I-V are from a female patient of 74. They show the typical subtrochanteric fissure extending transversely across varying widths of the femoral shaft. The changes over a period of four months are well seen in the serial X-rays, and it requires little imagination to visualize this fissure becoming the complete subtrochanteric break so commonly seen in fracture in Paget's disease of the femur. In the above patient, when weight bearing commenced, the edges of the fracture smoothed off, and the medial segment of the fissure appeared to be healed.

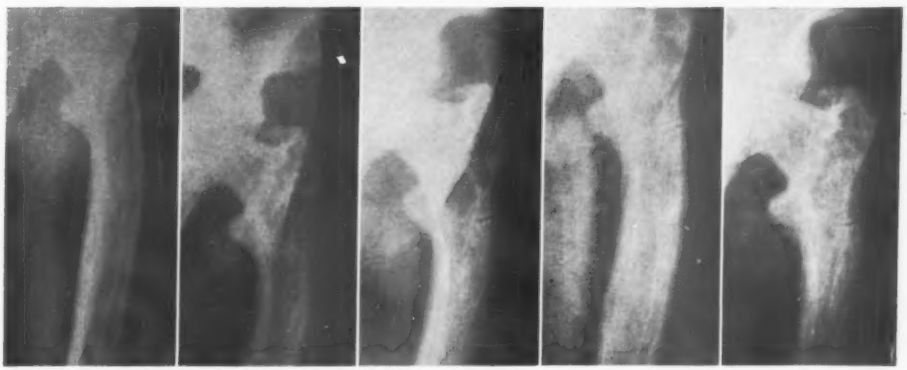

FIG. I FIG. II FIG. III FIG. IV FIG. V.

FIG. I. 5th April, 1953.—No obvious fissure present in the subtrochanteric region, but there is an area of transradiancy in the lateral medulla at this level. There is well marked periosteal bone deposition along the whole medial margin of the femur.

FIG. II. 20th April, 1953.—There is a complaint of pain below the left hip present for two days. X-ray shows a fissure fracture at the subtrochanteric level extending transversely across half the width of the femur.

FIG. III. 21st May, 1953.—There is definite sclerosis of the margins of the fracture with irregular resorption between these margins.

FIG. IV. 14th July, 1953.—The patient is now weight bearing. The edges of the fracture in the X-ray have changed their irregular shape and the sclerosis is less obvious.

FIG. V. 6th August, 1953.—There is healing of the medial segmont of the fissure. Sclerosis is absent. The outer margin of the femur is still interrupted by he original fissure.

ANATOMY OF THE FRACTURE IN PAGET'S DISEASE

Partial fractures

The fracture may be complete or incomplete. Allen and John (1937) and Rogers and Ulin (1936) have regarded incomplete or fissure fractures as being pseudo-fractures, i.e., not related to trauma or stress. Their occurrence on the convex margin of a bowed bone and the observation of a fissure becoming a complete transverse break, make this conception unlikely.

Partial fractures are common in Paget bone, particularly in the transition from vascular to sclerotic phases of the disease in long bones. They are said to be painful, but this is only true if the fissure is extending, or if it is sufficiently wide to permit movement at the site. Many fissures studied in patients did not appear to cause any symptoms clinically.

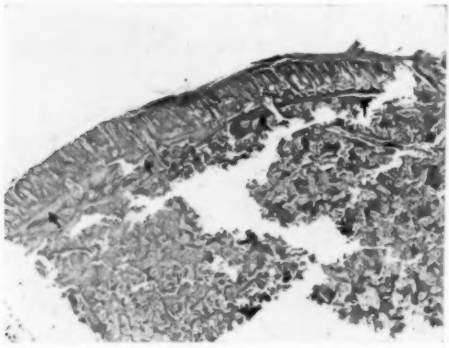

FIG. VI. Section showing partial fracture in Paget's disease. Note also the vessel showing endarteritis obliterans on the periosteal surface.

Sections of partial fractures are very difficult to acquire. Fig. VI is spoiled by artefacts, but several important points are demonstrated by the section. It shows the general direction of the trabeculae of the affected bone running in their usual line. A cleft has appeared in a bar of bone which crosses this direction transversely. This cleft runs across most of the section. These clefts were noted twenty years ago by Schmorl (1930), and it has been suggested that they are artefacts produced by the process of making the section. They are most uncommon, however, in sections of normal bone. It could be that the cohesion between successive areas of ossification is much weaker in Paget bone, and with the short and brittle trabeculae, are responsible for the appearance of fissures.

There may be another reason. The proliferative nature of the Paget change is marked in some bones. This, plus compression forces, could produce the typical cracks on the convexity of bowed bones. Such fissures in the concave aspect of deformed bones are seen in non-proliferative bone dystrophies, for example in fragilitas ossium.

The clinical inference from the microscopic section is that once the bone texture and mechanical stress produce a cleft, the remodelling process may perpetuate the error, thus increasing the likelihood of a complete fracture. Clinical experience supports such a suggestion, in that most of the fractures occurring in a Paget area are transverse. Furthermore, the experience of the elderly female patient mentioned above suggests that it is extremely difficult to completely cure such a fissure once it has extended an appreciable distance into the bone, even though Schmorl (1932) found the zones to be filled with osseous tissue.

TABLE 1

COMPLETE FRACTURE OCCURRING AMONG 255 ADMISSIONS WITH PAGET'S DISEASE

Femur, neck and subtrochanteric	25
Femur, shaft	16
Tibia	9
Humerus	8
Spine	7
Pelvis	11
Patella	1
Mandible	1
	78

Complete fractures

Complete fractures in Paget bone are usually transverse. In the majority of the series published by Lake (1951) where the fracture was not transverse it was doubtful whether the break had occurred in a Paget area. The short and irregular longitudinal bars are obviously less able to withstand transverse forces than the normal long trabeculae, not to mention the presence of osteoid tissue and vascular fibrous change as weakening factors.

The sites of complete fracture in Paget bone are shown in the Table. This is derived from a study of 255 admissions with Paget's disease to three Australian teaching

hospitals over five-year periods. The series of 78 fractures shows that the femur, tibia, humerus, spine and pelvis were the most frequently involved. This is in substantial agreement with other published smaller series (Brailsford, 1938; Gutman, 1936; Lennox, 1949; Rogers, 1936, and Wood, 1939) excepting that from the Mayo Clinic (Dickson, 1945) where the incidence of spinal fractures and cord compression was extremely high. This could well be a manifestation of selective hospital admission. In this present series there were six re-fractures in the same bone and on one occasion, three fractures occurred successively in the same bone. Fractures of the following bones in Paget's disease are apparently very uncommon. They are skull, maxilla, scapula, wrist, hand, foot and toe bones.

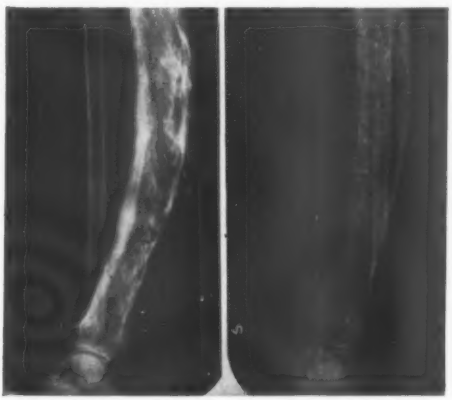

FIG. VII. Presenting with pain in the lower shin. The antero-posterior view (26th July, 1952) shows transverse fracture mid and lower third tibia without displacement. Lateral bowing of the tibia marked. The lateral view shows fracture is through two-thirds of the bone width. There are two fissures, the longer one running down and across from the crack which involves the anterior margin. Note that there is a completely normal fibula. It is straight and does not appear to be radiologically involved by Paget's disease.

The nature of the violence producing the fracture has been discussed by Allen and John (1937) and Rogers and Ulin (1936). This present study suggests that spontaneous fractures are less common than is generally supposed. In this connection a spontaneous fracture is defined as occurring without recognized violence or stress, or with minimal violence. A pathological fracture is one occurring in a pathological bone and the violence may be severe. Where the history was reliable (64 fractures), the trauma was moderate or severe in about half the cases (31 fractures).

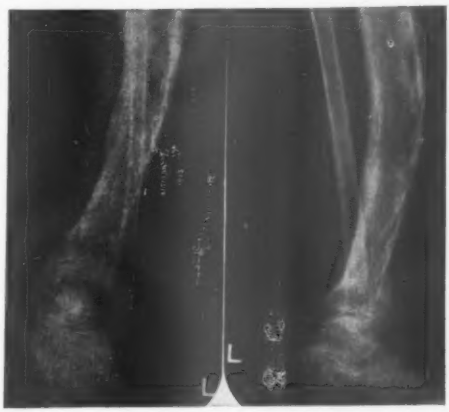

FIG. VIII. On 5th September, 1952, the fracture line is still present, healing has occurred over more than half of the fracture length. In the lateral view the inferior fissure is healed and there appeared to have been fibrosis at least, of the superior fissure.

In only 9 patients was there absolutely no force of any kind recognized. Many of the pathological fractures in the series suffered extreme violence. This problem of spontaneous and pathological fractures is not an academic one, as it is believed that the sclerotic bone of Paget's disease is often moderately strong, when the process has become inactive. Before this stage is reached honeycombing and vascularity will lessen such strength, and increase the likelihood of minor violence producing the fracture. Fracture was found to be commoner in the vascular stage (Dickson, 1945; Lake, 1951, and Wood, 1939).

It is believed that the determining factor in the production of most spontaneous fractures in Paget's disease, all other factors being equal, is the presence or absence of osteoporosis in the affected bone. Many patients in the Paget age group also have osteoporosis. This is the product of age, hormonal change, and disuse. It results in resorption, and inadequate deposition of protein matrix, and weakening of bone. It is believed when such changes are added to the

vascular or partially healed stage of the Paget process, a spontaneous fracture becomes quite probable. Recognition of this osteoporotic factor will aid the treatment of fractures in the Paget's disease, and is the subject of another paper (Lake, 1957).

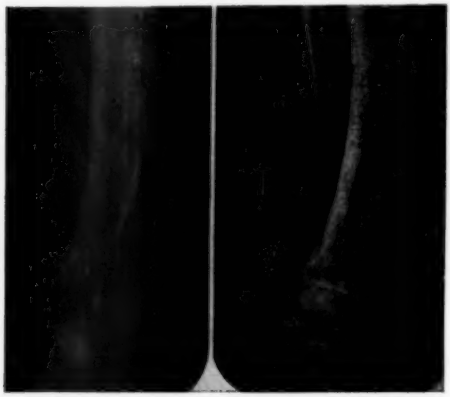

FIG. IX. Pain in the lower tibia has recurred (5th February, 1954). Antero-posterior: Probable partial fracture in the old site, hidden by fibula. Lateral: There is a linear rarefaction of the anterior margin on the old fracture site.

REPAIR OF THE FRACTURE

In the present series, about one-third of patients with long bone fractures (22), united at a faster rate than that calculated for a normal bone, and in a few lower-limb cases the time for clinical union was decreased by six weeks or more. One patient with a transverse fracture of the shaft of the femur was clinically and radiologically consolidated in three months. Conversely delayed or absent union was too frequent, being present in 14 patients, all with fractures of long bones.

Callus occurring in Paget fractures has several unusual features. It is occasionally quite abundant, but never as much as the "hyperplastic callus" seen in the rare case of fragilitas ossium. I did suggest (1951) that the callus was more likely to be abundant in the vascular stage of Paget's disease, but it is now considered that another factor may be operating. It is generally accepted that the increase of movement at the fracture site will produce an increase in the amount of callus and cartilage. It is now believed that this is the principal reason for any excess of callus seen in Paget fractures. Where there is no movement possible, e.g. in some small partial fractures, callus is often very scanty or even absent.

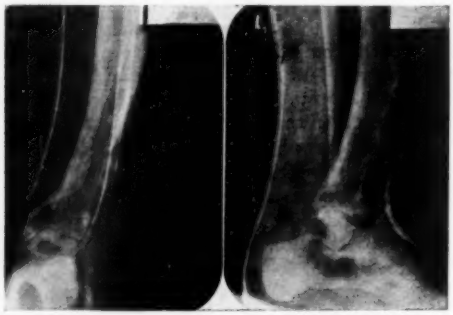

FIG. X. 29th March, 1954.—Antero-posterior: Irregular fracture site visible through light bandage. Lateral: Irregular fracture line in old site now more directly transverse. There is an inferior limb present.

Jaffe (1933) has described the callus and remodelling as participating in the Paget process. It has not been possible to obtain a section of healed Paget fracture, but where sections have been taken of partially healed callus at operation, they reveal an excess of cartilage where the callus has been abundant.

It is difficult to say with certainty just how important the vascular phase is in the production of excessive callus, but most of the cases of delayed or absent union occurred in patients with very sclerotic bone. This would appear to bear some relation to the vascularity of such bone, but many of these fractures in the past have suuffered some neglect by the medical attendant because of the associated bone dystrophy.

Such difficulty in healing is well illustrated in the case of a female aged 68 (Figs. VII-XI). Over a three-year period there were three complete transverse fractures of the lower tibia at the same site. Healing appeared to have occurred with appropriate treatment, after each episode, but re-fracture followed within a few weeks of activity. Fractures at this site in ordinary bones have a bad reputation, but on each occasion consolidation appeared to be complete. The remodelling of the healing area appears to have

prevented adequate cross trabeculation at the fracture site. This conclusion is in contradiction to the statement expressed by Brailsford (1938) in his excellent review. He stated that fracture at the same site is unusual owing to the efficiency of callus formation. This is generally true, but in this patient it was obviously not the case.

The observation of Fairbank (1950) that decalcification is common in the fractured region may be supported and his warning that neoplasm may be suspect from this change is echoed. The only method of assessing the problem is by serial X-rays and phosphatase determinations (Lake, 1951).

On the other hand, sclerosis is occasionally seen to follow fracture and it maybe that the fracture and the associated violence have accelerated the change from vascular to sclerotic stages.

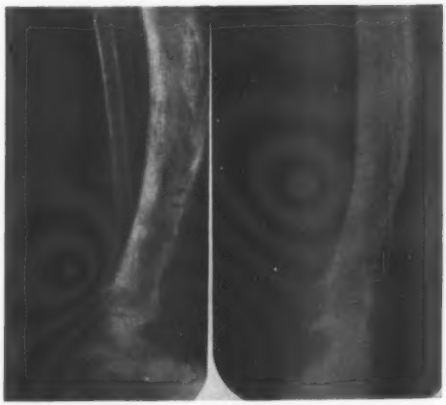

FIG. XI. 15th May, 1954.—Antero-posterior: Fracture line quite obvious in the mid and lower third of the tibia. Lateral: Healing is in progress.

Regarding the excessive callus and occasional astonishing rapidity of union, it may be that there is a hormonal or vascular effect on the repair processes. It is felt however, that slight movement with increase in the amount of callus for this reason, does aid union. The combination of all these possibilities would be comparable to the speed of union to be observed in most children suspended by gallows traction for fractured shaft of the femur.

SUMMARY

1. The anatomy and repair of fracture in Paget's disease is described.

2. The fracture may be partial or complete. Fractures are more common in the vascular phase.

3. Partial fractures are probably the result of local stress. The proliferative nature of the Paget process, plus lack of cohesion of successive osteoid layers appear to be responsible for fissure fractures. Healing of these is retarded by remodelling.

4. Complete fractures are usually transverse if occurring in a Paget area. The usual fracture sites are tabulated. Re-fracture is not uncommon.

5. Trauma producing such fractures is appreciable in about half the cases.

6. Osteoporosis is believed to be a determining factor in many fractures. That is to say that two bone dystrophies may be present in the aged patient.

7. There was rapid union in one-third of long bone fractures, and conversely there was serious delay in others.

8. Vascularity and hormonal factors may be important, but slight or marked movement at the fracture site may be the main influence on the speed of union.

ACKNOWLEDGEMENT

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MANAGEMENT OF NON-TUBERCULOUS THORACIC EMPYEMA

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Introduction

EMPYEMA of the pleural cavity has shown an increase in incidence in the past two to three years. Following the introduction of first the sulphonamides, and then penicillin in the treatment of pneumonia, pleural complications became very uncommon. I think this is partly the reason for the recent increased incidence. There is a need to relearn some basic principles, and I feel there is merit in reviewing the present management of non-tuberculous thoracic empyema. I propose to discuss briefly the pathology and treatment and illustrate these with selected cases from a total of 32 patients who have been referred during the past five years. All but five of these presented in the past two years. The discussion will not include post-operative pleural infections, but the management of empyema following thoracotomy and upper abdominal operations is similar.

Pathology

An empyema is a collection of pus in the pleural cavity. When an acute infection of the pleura proceeds to suppuration, the pus becomes loculated by fibrinous adhesions unless the infection involves the whole pleural cavity. With progression of this process a mature abscess is present. The walls of the abscess are formed by several layers. That closest to the collection of pus is a thick layer of fibrin often forming irregular masses. Outside this is a layer of granulation tissue and beyond this, on the surface of the pleura is a layer of fibrous tissue of varying thickness. As the process continues, more and more of the inner fibrin layer becomes organized on its deep surface, so that the thickness of the fibrous layer increases. The fibrinous and fibrous layers lining the pleurae are often referred to as the cortex. This becomes intimately connected to the parietal pleura except, in the early stages, over the diaphragm, so that a satisfactory plane of dissection cannot be obtained between the two. However, the cortex lining the visceral pleura only becomes intimately attached, usually temporarily, to pleura adjacent to areas of acute lung inflammation. Elsewhere a satisfactory plane of dissection is present (Fig. III). I believe the reason for this difference in the attachment of cortex to the parietal and visceral pleurae, is due to the elasticity of the underlying lung which retains more movement than the thoracic wall. The natural tendency of fibrous tissue to contract results in deformity of the involved hemithorax — approximation of ribs leading to scoliosis, fixation of the diaphragm and of the underlying lung. The extent of deformity will depend on the extent of the empyema.

I have discussed the pathology in the untreated case. The use of antibiotics to treat an underlying pneumonia alters not only the clinical course but also to some extent the pathological processes. Parenteral penicillin does not enter the pleural lesion in significant concentration but the broad-spectrum antibiotics do. Except in staphylococcal empyema the contents are usually rendered sterile, often prior to the first aspiration. This does not, however, result in the resolution of the pleural inflammation but rather its earlier maturation, so that a thick layer of fibrous tissue lines the pleura only three to four weeks after the onset of the infection. Further, the fluid may not look like pus on diagnostic aspiration which may yield thin, faintly turbid, yellow fluid — this is supernatant fluid; a thick purulent deposit lies at the bottom of the empyema cavity.

When does an empyema become chronic? Some consider that a definite number of weeks from onset should determine chronicity, but the progress of the pathological processes varies with the organism and the patient and the use of antibiotics. Others base the definition on the pathological state of the lesion. Barrett (1954) defines an acute empyema as one in which the pleura is lined by infected fibrin whereas in a chronic empyema the

fibrin has changed to fibrous tissue. This is unsatisfactory as we have no way of knowing the nature of the lining material until it is excised and submitted to microscopy. Fibrous tissue may be formed within two to three weeks of the onset of the process. I think it better to consider an empyema chronic when lung expansion is stationary, i.e. progress in the obliteration of the empyema cavity has ceased. However, I intend to avoid the use of the term and to consider the management of empyema based on the foregoing pathology.

MANAGEMENT

We must first consider the aims of our treatment. These must be the sterilization of the pleural cavity and the return of the hemithorax to normal, i.e. the complete expansion of the lung and the restoration of the normal movements of the chest wall and diaphragm.

In order to discuss the management of empyema, it is necessary to consider the treatment of acute infection of the pleura. The first essential is early recognition of a pleural complication of pulmonary inflammation. Lack of clinical response to treatment, or deterioration, or relapse in the patient's condition, should lead one to suspect a pleural lesion. Radiological observation of pneumonia is important. Any large or persistent pleural opacity should be investigated. Patients have been known to be discharged from hospital having recovered from pneumonia only to be re-admitted later with an empyema.

A recent example of this was a patient who had had a large interlobar effusion associated with a posterior basal pneumonia. His radiographs showed rapid clearing and absorption of the effusion with antibiotic treatment. A rather indefinite small, posterior basal opacity was still present in the lateral radiograph only, but he was well. A pleural rub, however, persisted. Radiographs a week later showed a more extensive, dense pleural opacity — in fact, there had now developed a loculated posterior basal empyema which has been treated.

Aspiration therapy

Once it is established that there is infected fluid in the pleural cavity, the aim must be to keep the cavity empty by aspiration followed by the instillation of the appropriate antibiotic. This is done twice daily if necessary and the frequency and site controlled by radiography. Accurate localization is essential. Penicillin 500,000 units in 5 mls. solution, should be used if the causative organism is a pneumococcus or an anaerobic streptococcus. If it is a staphylococcal infection, then the antibiotic to be used — one of the tetracycline group, chloramphenicol or possibly erythromycin — is determined by sensitivity tests. The best form of the former for intrapleural use is the intravenous (500 mg.) or intramuscular (400 mg.) preparation. Chloramphenicol (500 mg.) may be introduced as a suspension. It has been incorrectly stated that, except in the case of penicillin and streptomycin, it is not possible to administer any of these antibiotics intrapleurally (Belcher and Grant, 1955). It is unnecessary to give the drug other than intrapleurally unless there is a lesion in the underlying lung which requires treatment. There is usually sufficient absorption of the drug from the pleural cavity to give an adequate blood level for the treatment of any pulmonary infection. Successful treatment is indicated by improvement in the clinical condition, but this can be misleading as regards the state of the thoracic lesion. Radiography should show progressive re-expansion of the lung.

Case report

E.R., a child of five years of age, was admitted to a hospital with left-sided pleuritic pain. Her radiograph was considered normal. A few days later a left opacity was present. This was treated with the tetracycline group, chloramphenicol, erythromycin and albamycin by mouth, in turn, over the next two weeks. This had no effect on her temperature or general condition. Aspiration was then attempted but only a few mls. of pus were obtained which gave a growth of staphylococcus aureus. She was then transferred to Wellington Hospital. A radiograph following admission showed a total empyema (Fig. I). Her temperature was 104°F and she looked ill. Aspiration under rectal thiopentone narcosis with local procaine 1 per cent. solution yielded 200 mls. of thick pus. Culture gave a pure growth of staphylococcus aureus. Two hours after aspiration her temperature was normal and remained so. No antibiotics were administered after admission. Aspirations were continued but at the end of each difficulty was experienced in finding the pleural cavity so no antibiotic was instilled. A radiograph one week after admission showed a lateral pleural opacity only (Fig. II). She was discharged three weeks later.

This case illustrates that antibiotics alone will not treat an empyema. The temperature

did not respond to antibiotic therapy although several were used, but it did respond dramatically to simple aspiration. Organisms could still be grown from the fluid removed at the last aspiration yet this treatment alone sufficed.

If the fluid is thick and some difficulty is experienced in aspirating it, a streptokinase-streptodornase solution of 200,000 units and 50,000 units respectively should be instilled. These enzymes have a fibrinolytic activity which may continue over a period of twenty-four to forty-eight hours. The introduction of this solution frequently causes a pyrexia which will respond to aspiration. To be effective this solution must be used early, probably in the first ten days.

If, after two weeks of this regime, the progress is inadequate then some surgical procedure is necessary. Most commonly the need for this arises because of, either inadequate, or failure of, aspiration treatment. Unsuspected disease in the underlying lung—tuberculosis, actinomycosis, ruptured hydatid cyst, lung slough following abscess formation or neoplasm, are less common causes of chronicity. Large bilateral empyemata may require surgical drainage earlier.

The following surgical procedures are available:—

1. Closed intercostal drainage;
2. rib resection with large tube drainage;
3. excision of the empyema.

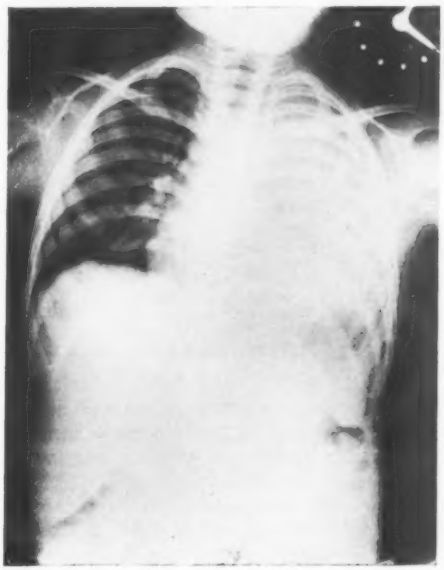

FIG. I. Radiograph of child aged 5 years on admission. A total left empyema is present with displacement of the mediastinum to the right.

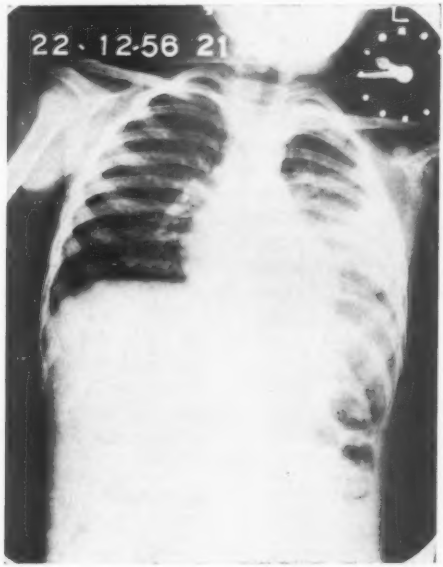

FIG. II. Radiograph of the same child as Fig. I., one week later after several aspirations had been undertaken.

Throughout this treatment, breathing and postural exercises should be practised and the patient mobilized. This will lessen the danger of thoracic wall contraction and encourage the absorption of fluid and lung re-expansion. As soon as toxaemia is overcome the patient should be ambulatory. Any anaemia should be corrected.

Surgical drainage

Closed intercostal drainage is the treatment of choice in the very young and the elderly. It is continuous drainage as opposed to intermittent drainage by aspiration. This method has special application in babies and infants with an empyema resulting from the rupture of a pneumatocoele or cyst associated with

staphylococcal pneumonia. It may be applied to adult patients who have a small loculated empyema which has failed to respond to aspiration therapy. Suction can be used as required. The main disadvantage in the past has been the blockage of the tube by fibrinous masses.

The advantages of rib resection over intercostal drainage are that these fibrinous masses can be removed at the operation and a larger bore tube utilized for drainage. The size of an intercostal tube is limited by the space between the ribs. However, the use of a streptokinase-streptodornase solution can ensure its patency. These enzymes not only thin the pus but the masses of fibrin are broken up so the objections to intercostal drainage are overcome. The enzymes are injected down the tube together with the appropriate antibiotic and the tube then clamped for a few hours. After its release, closed drainage is continued. This must not be done in the presence of a fistula.

A further advantage of closed intercostal drainage is that the pus drains into a bottle containing an antiseptic and no dressings are required. This reduces the possibility of cross-infection and saves nurses' time. There is no necessity for the patient to remain in bed. It is important that the tube be placed so as to give dependant drainage. The instillation of a radio-opaque iodine preparation followed by radiographs in the postero-anterior and lateral planes will provide the necessary information. The tube is removed when sinograms demonstrate no pleural space, usually within two to three weeks from the commencement of drainage.

The indications then, for closed intercostal drainage are:—

1. Empyema in babies and infants and the elderly, and
2. a small loculated empyema in children and adults.

With the management outlined above rib resection seems unnecessary. All other empyemata which do not respond to aspiration treatment are best dealt with by the excision of the empyema.

Excision of the empyema

Experience with clotted and infected haemothoraces in the latter part of World War II showed that early pulmonary decortication gave gratifying results both in overcoming the infection and restoring pulmonary function.

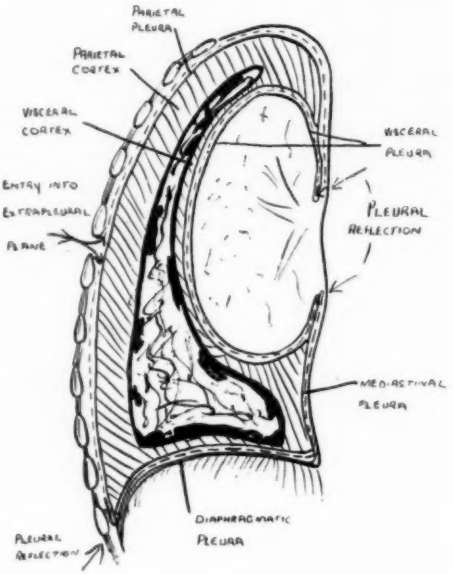

FIG. III. Diagrammatic representation of a total empyema showing the plane of dissection followed in the operation of empyema excision.

In 1946, Sanger applied this experience to a young soldier who, as a result of a pneumonia, developed a multilocular empyema. Ten days from the onset of his illness, aspiration was performed and after a further three days two intercostal catheters were inserted. As the catheters ceased to drain after three days, thoractomy was undertaken five days following the institution of catheter drainage. The operation he practised was to open the pleural cavity, remove all fibrin clots and debris and then decorticate the lung. He stated that this operation should be done early —that is, two to three weeks from the onset, but did not consider it was necessary to deal with the parietal cortex. In 1947, Brock suggested that primary decortication was indicated in the presence of a large empyema and, in 1948, practised pulmonary decortication with partial parietal decortication two and a half weeks following the onset of an effusion

associated with a pneumonia (Fry, 1948). In subsequent years, it was recognized that excision of the parietal pleura and its densely adherent cortex, which is thicker than the visceral cortex, was necessary in order to restore function of the diaphragm and chest wall. The operation then adopted was excision of the empyema mobilizing the sac by extrapleural dissection over the parietes. This extrapleural mobilization had been practised by Fowler in 1901, but it was not until it was developed by Sarot in 1949, principally for tuberculous empyema, that it came into general use.

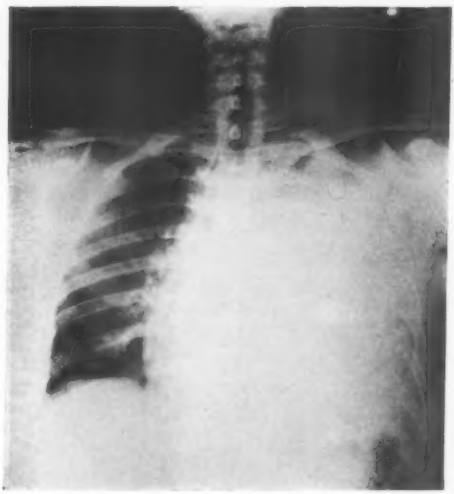

FIG. IV. Radiograph of male patient aged 26 years on admission. There is a total left empyema with displacement of the mediastinum to the right.

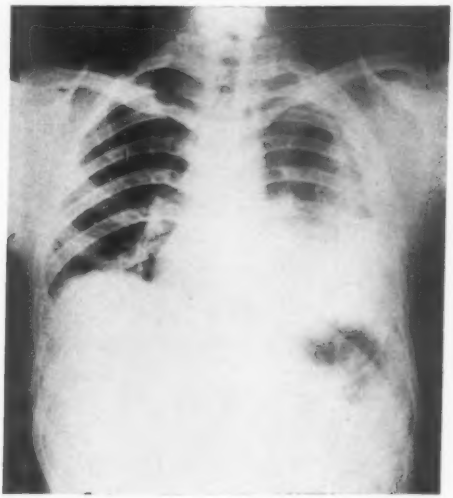

FIG. V. The same patient two weeks later following frequent aspiration. The air in the pleural cavity shows well the thickness of the visceral and parietal layers of cortex.

Pulmonary decortication was practised many years ago for late empyema which had been present for months or years. The proper place of empyema excision is in the management of a large acute empyema particularly in childhood, and multilocular or total empyema.

Through a standard postero-lateral thoracotomy dividing but not resecting a rib, the empyema sac and its contents are excised as a whole by mobilization of the sac in the extrapleural plane over the costal parietes and decortication of the lung and diaphragm (Fig. III). Infolding of the lung, due to adhesions developing when it was collapsed by the empyema, must be dealt with by further mobilization after decortication. Confusion exists regarding the terminology of the operation described above as excision of the empyema. Decortication refers to excision of cortex but the costal parietal pleura is also removed. Pleurectomy is often used to describe the above operation but the visceral and diaphragmatic and frequently the mediastinal pleurae are left intact.

If this operation is performed within four weeks from the onset of pleural infection, mobilization is easy; at six or seven weeks, the adherence of the costal parietal pleura and cortex may be considerable. The vascularity here is increased and haemorrhage can be substantial. For this reason, all the cases which have undergone this operation have had hypotensive anaesthesia. As swab-weighing is routine practice, a fairly accurate estimate of blood loss is possible and blood is replaced as loss occurs. The average blood loss in the later cases is approximately 1,000 mls. Two or three intercostal catheters size 21 Fr. are inserted, usually in apical, basal

and anterior positions. Suction is applied as long as air leaks or drainage continue, usually twenty-four to forty-eight hours. The patient is mobilized early, primary healing occurs and he can usually be discharged two to four weeks after operation. This contrasts with the protracted convalescence following rib resection after which a drainage tube may remain for weeks or months and seldom less than six to eight weeks. Another considerable advantage is the removal of the source of toxaemia at the time of the excision operation. Further, any pulmonary lesion requiring surgical treatment can be resected.

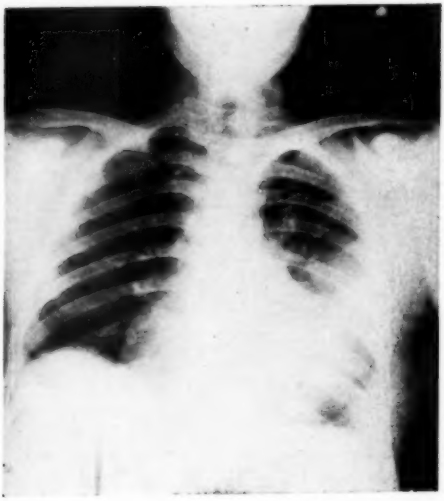

FIG. VI. Radiograph of the same patient one week after excision of the empyema.

Case report

D.P., a male aged 26 years, was admitted to hospital with a three-day history of left chest pain, breathlessness and loss of appetite. He was pale, and breathless sitting in bed. There was reduced movement and dullness over the left chest where air entry was present only over the upper chest anteriorly. A radiograph showed a generalized left opacity with displacement of the mediastinum to the right (Fig. IV). Fluid removed by diagnostic aspiration was clear, yellow and odourless and gave no growth on culture for forty-eight hours. Further aspirations were not undertaken for two days. Over the course of the next ten days 4½ litres of fluid were aspirated. The fluid was becoming thicker and more difficult to remove. Cultures were sterile. A radiograph at this stage showed a long narrow pleural air space revealing a cortex on the visceral pleura 1 cm. thick and on the parietes, 1.5 cm. thick. A large basal opacity was still present (Fig. V).

During the subsequent nine days, aspirations yielded a total of 2 litres of purulent fluid. After each aspiration, a streptokinase-streptodornase solution had been instilled. He was then transferred to the Thoracic Surgery Unit, and three days later (three and one-half weeks from the onset of his pleural symptoms) he was submitted to thoracotomy and empyema excision. Hypotensive anaesthesia was employed by Dr. V. B. Cook. Blood loss was 250 mls.

FIG. VII shows the empyema sac from the anterior aspect, and Fig. VIII the contents and the thickness of the wall. Histologically, there was a considerable amount of granulation tissue present. The content was thick grey purulent debris.

The post-operative course was uneventful. The drainage tubes were removed on the second day and aspiration of 350 mls. of thin straw-coloured pleural fluid was performed in the mid-axilla. He became ambulant the same day. A radiograph one week after operation (Fig. VI) showed satisfactory appearances and he was discharged three weeks after operation. Movements of the chest wall were normal.

This case history illustrates the course followed in patients who are submitted to excision of the empyema at the optimum time.

DISCUSSION AND RESULTS

During the five-year period 1948-1952, eleven patients were admitted and treated for empyema. This contrasts with 32 patients who have been referred over the years 1953-57, inclusive. Since the establishment of this Unit five years ago patients have been referred from other hospitals. These patients account for approximately half of those observed. In some of the recent cases there is no doubt that the pleural lesion had not been recognized at first. The widespread use of antibiotics has resulted in the production of sterile empyemata which require just as energetic treatment as those in which organisms remain. The pleural lesion requires local treatment as has been indicated.

Another reason for the increased incidence of empyemata is the occurrence of antibiotic-resistant staphylococcal infections. In this series, all the cases due to the latter organism have presented in the past eighteen months. Table 1 gives the bacteriology of the fluid at the time of the first aspiration. Of the seven sterile empyemata, one was probably staphylococcal and two streptococcal.

TABLE 1

BACTERIOLOGY

Causative Organism	*Number*
Staphylococcus aureus	14
Pneumococcus	6
Anaerobic streptococcus	3
B. coli	1
B. pyocyaneus	1
Sterile	7
Total:	32

FIG. VII. Photograph of the empyema sac from the anterior aspect.

infant, three adults with small empyemata and the seventh an elderly woman. Only two had a tube for approximately three weeks; in the others it was removed in less than two weeks.

TABLE 2

METHOD OF TREATMENT

Aspiration alone	10
Intercostal tube	7
Rib resection	1
Excision of empyema	14
Total:	32

FIG. VIII. The sac has been opened to demonstrate the contents and the thickness of its wall.

Table 2 gives the method of treatment employed. The rib resection was done four years ago for a large posterior basal empyema in a patient who refused a more major procedure. The result was entirely satisfactory but drainage was required for eight weeks. Of the seven patients who were treated by intercostal drainage, two were babies, one an

Empyema excision

Two patients were under ten years of age, the youngest being 3 years and two were over 50 years; five were in the third decade and the remaining five in the fourth and fifth decades. There were ten males and four females.

There was no operative or late mortality.

The post-operative complications were few. One patient had a major pulmonary embolus from which he made a complete recovery. Another developed a local wound abscess — she was a major isolation problem as she was a nasal carrier of epidemic type staphylococcus (phage type 80/81). A third had a small residual empyema pocket which responded to drainage. He was a late case, his operation being about ten weeks after the onset of his pleural lesion. There were no other complications.

It has been stated that this operation should not be practised in less than six to eight weeks from the onset of pleural infection and that the disadvantages of the operation are the possibility of a residual empyema pocket requiring drainage and the recrudescence of infection (Holmes Sellors and Cruickshank, 1951).

Neither of these complications are likely if the operation is performed within four weeks of the occurrence of pleural suppuration. At this stage the tissues are still mobile and space obliteration occurs rapidly after excision. Early operation avoids the necessity for multilating flap operations on the chest wall to close infected dead spaces associated with late empyema. Further, blood loss is less with early operation.

It has been maintained that because these are severe procedures they should be reserved for selected cases such as total empyema. It cannot be denied that excision is a much more major operation than rib resection but it has been shown that it is safe and that the convalescence is very much shorter.

SUMMARY AND CONCLUSIONS

Thoracic empyema is increasing in incidence. Of 32 cases referred for management over a five-year period, 27 have occurred in the past two years. Part of this increase is due to antibiotic-resistant staphylococcal empyema.

The treatment of pleural suppuration must be vigorous consisting of frequent aspirations of the fluid with the instillation of the appropriate antibiotic and proteolytic enzymes as required. The aim must be to render the pleural cavity dry and sterile, and restore normal function to the thorax.

Thoracic physiotherapy must be instituted early.

Radiographic control is essential. Failure to obtain progressive re-expansion of the lung is an indication for surgical treatment.

Intercostal catheter drainage is the treatment of choice in babies, infants and the elderly and in small loculated empyemata. All other empyemata are best treated by excision. This has been practised in 14 patients without mortality. The advantages are:—

1. Primary healing.
2. The patient is fit for discharge 2-3 weeks post-operatively.
3. The source of toxaemia is immediately removed.

The optimum time for operation is under four weeks from the onset of pleural infection.

ACKNOWLEDGEMENTS

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